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LUNG FUNCTION STUDIES

VII. THE EFFECT OF PNEUMONECTOMY ON RESPIRATORY DEAD SPACE

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WITH pneumonectomy, part of the bronchial tree is removed. However, there is disagreement about the effect of pneumonectomy on the volume of the respiratory dead space. It has been reported that, after pneumonectomy, the dead space is enlarged;[1, 2] or that it is decreased.[3, 4] The former situation, if present, would be a considerable factor contributing to respiratory inefficiency.

The development by Lilly and Hervey of the N_2 meter, for continuous analysis of the N_2 concentration of respired gases, has made possible the ready measurement of respiratory dead space both of normal subjects[5] and of many patients with abnormally uneven alveolar ventilation.[6] This measurement of respiratory dead space represents the volume of inspired gas which remains in the conducting airway and does not contribute to dilution of alveolar gas. It probably approximates the so-called anatomic dead space, and is practically independent of alveolar ventilation-perfusion relationships, which affect other types of "dead space" measurement using oxygen or carbon dioxide values.[7]

Measurements of the respiratory dead space in patients before and after pneumonectomy are presented; the postoperative values were similar to those of healthy control subjects. Other studies made after pneumonectomy suggest that the large "dead space" values reported by Birath were caused by abnormally uneven alveolar ventilation, and not by an actual enlargement of the dead space.

METHODS

Eight patients (7 males and 1 female) were studied after pneumonectomy for pulmonary neoplasm; seven were also studied a few days preoperatively. Eleven elderly (53 to 71 years) male hospital patients without clinical or fluoro-

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scopic evidence of pulmonary disease served as controls. All subjects were examined semi-reclining in bed, with the head and trunk elevated 50 to 60 degrees. Subjects were not in basal condition but had rested in bed at least ten minutes before measurements were made.

The method has been described previously.[5, 6] In brief, it consists of continuous analysis and photographic recording of N_2 concentration and volume flow of gas expired during and after the change from breathing room air to breathing 99.6 per cent oxygen. Although the dead space of most young normal subjects can be measured directly on the photographic records,[5] it was necessary here to replot two or three successive expirations (occurring during the first thirty seconds of O_2 inhalation) as curves of N_2 concentration versus expired volume.[6]

Dead space measurements made on different days (before and after pneumonectomy) might differ because of error in the method and also because of day-to-day variation, quite independently of the operation. Therefore duplicate measurements were made on six healthy subjects on each of two different days. By analysis of variance[8, 9] of these measurements and of the duplicate measurements on the patients, it was found that (a) the error of the method was similar in the patients, the six healthy subjects noted above, and in forty-five healthy subjects studied previously,[5] and (b) a difference of 42 ml. or more between the means of duplicate pre- and postoperative measurements, made on different days, as given in Table I, is significant and may reasonably be ascribed to the operation.

The method requires the inhalation of O_2, which, over a five minute period, usually causes a reduction of tidal volume in chronically anoxemic patients, and yields a larger dead space/tidal volume fraction than if the average tidal volume observed during a five minute period of breathing air is used. The dead

TABLE I. RESPIRATORY DEAD SPACE BEFORE AND AFTER PNEUMONECTOMY,* AND IN NORMAL ELDERLY MALES

	SEX	AGE YRS.	HT. CM.	RESP. D. S. ML.		TIDAL VOL. ML.	DS/TV X 100 %	INTERVAL	PNEUMONECTOMY
Normal†	M	59.6	166	150		623	24.9	----	----
Mean ± S.D.				±31		±149	±6.1		
				pre	postoperative				
Na	M	60	171	187	133	494	27.0	13 days	R
Ws	M	59	175	154	144	452	31.8	13 days	L
Fr	M	55	173	203	140	582	24.1	12 days	L
Wh	M	65	161	--	130	386	33.6	3 mos.	R
Gn	M	54	178	205	158	576	27.4	12 days	R
Cy	M	68	165	178	190	906	21.0	5 mos.	R
Ry	M	52	167	202	145	670	21.6	9 mos.	L
Mn	F	56	165	137	125	393	31.8	15 days	R
Mean (males)	–	59	169	188	149	581	26.6	----	--

*All male patients had carcinoma, *Mn* had mesothelioma; in addition, *Cy* had tuberculosis, silicosis and emphysema and *Ry* had emphysema.

†These 11 subjects were reported elsewhere.[6]

space/tidal volume fractions given (Table I) were calculated from average tidal volumes during the breathing of air, measured with a recording 120-liter spirometer. All volumes are corrected to BTPS.

RESULTS

Data for patients and control subjects are given in Table I. Preoperatively, the dead space values in men, 154 to 205 ml., fell in the upper range of normal values, 150 to 212 ml. (average plus 2 S.D.). The mean value of 188 ml. for the male patients was significantly ($p = 0.02$) greater than the mean control value of 150 millimeters.

Postoperatively there were no significant changes in the dead space values for two men and one woman. The lack of change in the woman may have been due to a dead space reduction also existing preoperatively, caused by compression stenosis of the right bronchial tree, including the right main bronchus, from the large tumor occupying about two-thirds of the right hemithorax. Four male subjects, none of whom had obstruction of main bronchi preoperatively, had significant postoperative reductions of about 50 millimeters.

The mean postoperative values of male patients for dead space and dead space/tidal volume fraction were similar to the values for the healthy males.

DISCUSSION

The explanation of the enlargement of the mean preoperative dead space value is not entirely clear. In the two cases with pulmonary emphysema, the values of 178 and 202 ml. probably reflect the over-all state of pulmonary hyperinflation exhibited during quiet breathing by such patients. A similar explanation may apply to some extent to the other cases; enlargement of the functional residual capacity has been reported in similar patients.[1]

The mean postoperative values for dead space and dead space/tidal volume fraction reported here are significantly smaller than the respective mean values of 236 ml. and 48.7 per cent reported by Birath,[1] which, with other similar findings,[2] were taken to indicate a probably actual enlargement of the dead space resulting from postoperative pulmonary distention. The difference very likely results from the methods used. Birath examined his patients in a sitting position, in which the respiratory dead space is about 25 ml. greater than when semi-reclining.[10] Probably even more important was the presence of uneven pulmonary ventilation in his subjects. In five of our patients studied postoperatively it was possible to make calculations similar to those used by Birath, since the alveolar (end-tidal) N_2 concentrations and tidal volume were measured continuously during the first two minutes of O_2 inhalation, and functional residual capacity was also measured. Four of these patients were found to have abnormally uneven ventilation by the method described elsewhere,[11] and their calculated "dead space" values were all larger, average 295 ml., than the direct measurements, average 152 ml.; the values were similar in one patient (Mn) with normal ventilation uniformity. The reasons for the discrepancy between the calculated and directly measured values follow. In Birath's calculations, based on rebreathing a gas mixture containing H_2, and in ours, which were based

on the pulmonary N_2 elimination equations of Darling and associates,[12] it is assumed that the lungs are ventilated uniformly, and that the ventilatory process can be expressed by an exponential equation, stating that the alveolar gas concentration is changed by a certain fraction on each successive inspiration of the "foreign gas." For example, if 300 ml. of O_2 (tidal volume of 450 ml. minus 150 ml. dead space) are added to the functional residual capacity of 2700 ml., containing 80 per cent N_2, the N_2 concentration will be reduced to $\frac{2700}{3000}$ or 0.9 times 80 per cent; the next breath will reduce the concentration to 0.9 times 72 per cent, etc. Experimentally one obtains the value of the fraction, and, knowing tidal volume and functional residual capacity, can solve for the dead space. However, with uneven ventilation, the hyperventilated areas of the lungs approach equilibrium with the inspired gas more rapidly (smaller value of the fraction) than do the relatively hypoventilated areas. Therefore, early in the procedure the expired gas will reflect the rapid rate of change in the hyperventilated areas more than it does later, when these areas have reached equilibrium and the expired gas is manifesting the slower rate of change in the hypoventilated areas. Such a multiphasic ventilation process cannot be expressed accurately by equations containing only one fractional value expressing the rate of change. One can calculate dead space using one fractional value, but the result varies with the portion of the equilibration curve from which the fraction was obtained. The rapid change early in the procedure yields a very small value for dead space, the slower change later in the procedure yields an excessively large value for dead space. Birath's calculations were apparently based principally on a measurement made after a one-minute breathing period; similar calculations applied to the one-minute point on the alveolar N_2 elimination curves gave the artificially large "dead space" values noted above.

The effect of pneumonectomy on the volume of the respiratory dead space is not easily anticipated; there appear to be various changes which could either diminish or enlarge it. The dead space, as measured here, represents the volume of inspired gas which remains in the conducting airway and does not contribute to dilution of alveolar gas. This volume is affected by the anatomic volume of the conducting airway and also by the extent of gaseous diffusion occurring between terminal bronchioles and alveolar spaces; prolongation of inspiratory time results in a smaller dead space, presumably because the peripheral boundary of pure inspired gas recedes up the bronchial tree.[5]

Partial resection of the bronchial tree would tend to reduce the dead space. Rohrer[13] estimated that about 45 per cent of the volume of the conducting airway was below the tracheal bifurcation. One might therefore expect a 20 to 25 per cent reduction with pneumonectomy; the mean change observed postoperatively was a 17 per cent reduction. However, part of the remaining airway is intrathoracic, and probably participates in any postoperative distention of the remaining lung. Also, the dead space might possibly be enlarged by a reduction in the extent of gaseous diffusion occurring between terminal bronchioles and alveolar spaces. This could result from shortened diffusion time accompanying a postoperative increase of respiratory frequency (an average in-

crease of 2.4 respirations per minute was noted in the patients above); a change in what may loosely be called diffusion surface might result when a total alveolar volume considerably greater than half normal is connected to bronchioles presumably only half normal in number.

A postoperative dead space volume of 150 ml., although similar to normal values, is in one sense a respiratory handicap. Normally only about 75 ml. of the tidal volume developed by each lung is not effective in ventilating the alveoli; thus the "wasted effort" of the remaining lung is doubled. This, in addition to other departures from the normal thoracic mechanics, represents an encroachment upon the reserve of the bellows or ventilatory mechanisms, and may contribute to the postoperative tendency to experience dyspnea. However, in another sense, a postoperative bronchial distention might be favorable. After pneumonectomy, the patient is obliged to force through one set of bronchial tubes about the same respiratory minute volume which is normally forced through two sets of tubes. The increase in respiratory effort needed for this would probably be reduced by bronchial distention in the remaining lung.

SUMMARY

The respiratory dead space was measured before and after pneumonectomy on patients breathing quietly in the semi-reclining position. Postoperatively, three subjects showed no change, and four male patients showed significant reductions. The mean postoperative values for dead space and dead space/tidal volume fraction were similar to control values for elderly men. Factors affecting the volume and measurement of respiratory dead space after pneumonectomy are discussed.

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OBSTRUCTIVE EMPHYSEMA WITH A DEFECT OF THE ANTERIOR MEDIASTINUM

Report of a Case

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OPENINGS in the anterior mediastinum with direct communication between two pleural cavities are very rare congenital abnormalities. Kupfer[1] studied mediastinal lesions in 6,000 autopsies and did not observe such an anomaly. In 1939, Oehsner, DeBakey, and Murray[2] reported a case of congenital absence of the anterior mediastinum visualized at operation for repair of a diaphragmatic hernia in an 18-month-old child. The entire anterior mediastinum was absent, and the right hemithorax could be visualized from the left thoracic wound. The pericardium and great vessels lacked their normal attachment to the posterior aspect of the sternum. The diaphragmatic hernia was repaired, but no repair of the mediastinum was carried out. The infant made an uneventful recovery.

In their report, a review of the literature revealed only nine other reports of human subjects suspected of having a passageway connecting the two pleural spaces. However, the report was the first clearly to describe the anatomic findings, which without a doubt, represented a congenital abnormality of the mediastinum.

Since that time a review of the literature reveals only one other case of proved congenital absence of the anterior mediastinum. In 1945, Gross and Lewis[3] reported an absent anterior mediastinum in a 4-year-old child who was operated upon for obstructive emphysema of the right upper lobe caused by absent cartilaginous rings in the right upper lobe bronchus. A right upper lobectomy was carried out, and the anterior mediastinum reconstructed by suturing the pericardium to the sternal periosteum with two rows of interrupted silk sutures. This was the first report of a successful surgical repair of the anterior mediastinum.

In view of the rarity of the defect, one of us (J. E. L.) feels rather fortunate in having been able to collaborate with Dr. Gross in reporting the previously proved case in 1945 and to add yet another case of anterior mediastinal defect associated with obstructive emphysema of the right middle lobe of the lung.

CASE REPORT

A 4-month-old white male infant was admitted to the Children's Memorial Hospital on the service of Dr. Stanley Gibson, June 1, 1950, with a history of dyspnea and cyanosis when crying noted since 6 weeks of age.

From the Surgical Service of the Children's Memorial Hospital, Chicago, Ill.
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Fig. 1.—Anteroposterior roentgenogram shows the heart pushed to the left, collapse of the right lower lobe, partial collapse of the right upper lobe, and marked emphysema of the middle lobe.

Fig. 2.—The lateral roentgenogram shows a large retrosternal radiotranslucent area. The heart is pushed posteriorly.

The infant had an uncomplicated spontaneous birth following a normal pregnancy. He weighed 5 pounds, 5 ounces at birth, and progressed normally in weight gain and development without associated symptoms until 6 weeks of age. At this time it was noted that the infant became blue and dyspneic when crying. However, there were no other symptoms, and the infant developed normally until six weeks before entry to the hospital. At this time he developed paroxysms of cough accompanied by crying, cyanosis, and rapid respirations. The attacks occurred approximately every half hour, and lasted from five to ten minutes. Associated with these attacks, he had difficulty in eating and lost weight. Feeding seemed to initiate difficulty in breathing and produced crying and cyanosis. Consequently he was brought to the hospital for evaluation and study.

Physical examination on admission revealed a rectal temperature of 104° F., a rapid pulse, and rapid respirations. The infant was well developed and well nourished, but slightly cyanotic. Respirations were grunting in character, punctuated by a dry hacking cough. Physical findings were essentially normal except for the chest examination. The anteroposterior diameter of the chest appeared prominent. The heart was shifted to the left, and the heart sounds were distant with no murmurs. There was a hyperresonance to percussion over the right anterior chest and over the cardiac area. Breath sounds were distant on the right side of the chest and normal on the left. No râles were heard.

Laboratory examination revealed a white blood count of 15,800, with 32 per cent polymorphonuclear leucocytes, 60 per cent lymphocytes, 5 per cent monocytes, 1 per cent eosinophiles, and 2 per cent stab forms; a hemoglobin of 10.4 grams per cent, and a red blood count of 4.42 million. The urine was normal.

Roentgenograms of the chest revealed emphysema of the right middle lobe with atelectasis of the right upper and lower lobes. The heart was shifted to the left. In the lateral view there was a clearly demarcated globular area of emphysema lying anterior to the heart beneath the sternum. The diagnostic impression was obstructive emphysema involving the right middle lobe of the lung (Figs. 1 and 2).

The infant's grave condition precluded further studies, and thoracotomy was carried out as an emergency procedure. The infant was anesthetized with endotracheal cyclopropane. A polyethylene tube was inserted in the right ankle vein for the administration of blood.

The infant's condition was very precarious under anesthesia because of the poor exchange of gases in the small amount of functioning lung, and thoracotomy was rapidly carried out through the fourth interspace. The large emphysematous middle lobe delivered itself partially through the operative wound, and the pulmonary difficulties were sufficiently relieved to allow deliberate procedure with the operation. Inspection revealed a large emphysematous middle lobe occupying the right chest cavity and collapsing the right upper and lower lobes. On withdrawing the remainder of the middle lobe from the chest cavity, it could be seen that the middle lobe extended across the midline anterior to the pericardium into the left hemithorax through a large anterior mediastinal defect. The left lung could be visualized without difficulty through the defect in the anterior mediastinum. Consequently a right middle lobectomy was carried out. The vessels were dissected out individually, ligated, and divided. The bronchus which appeared grossly normal was clamped with a toothed ductus clamp, divided and sutured with interrupted No. 00000 silk sutures. Pleura was then sutured over the stump of the bronchus. The question of closure of the anterior mediastinum then arose. It was felt desirable, but inadvisable, in view of the precarious state of the patient, the technical difficulty from the posterolateral approach, and the increased anteroposterior diameter of the chest which would have contributed marked tension to the suture line.

Consequently, the chest was closed in layers with a catheter inserted into the pleural cavity through the seventh interspace for drainage.

At the close of the operation the infant's condition seemed good; however, as the intratracheal catheter was removed, respirations ceased. The catheter was quickly reinserted and the patient revived with artificial respiration and oxygen. This sequence of

events took place three more times before normal respiration was reestablished. The infant was returned to his crib approximately one hour after surgery in good condition. His postoperative course was excellent (Fig. 3). Just prior to discharge on the tenth postoperative day, roentgenograms revealed that the heart had shifted back to its normal position, and there was complete expansion of the right upper and lower lobes (Fig. 4). No evidence of recurrent emphysema or residual atelectasis was seen. The infant was discharged without symptoms, and when seen recently at the age of 7 months was in excellent condition.

Pathologic review of the lobe revealed no cause for the emphysema. The bronchus, bronchi, and bronchioles appeared patent and normal. The lung presented a typical picture of emphysema. The main bronchus was sectioned, and revealed no cartilaginous ring defects or obstruction.

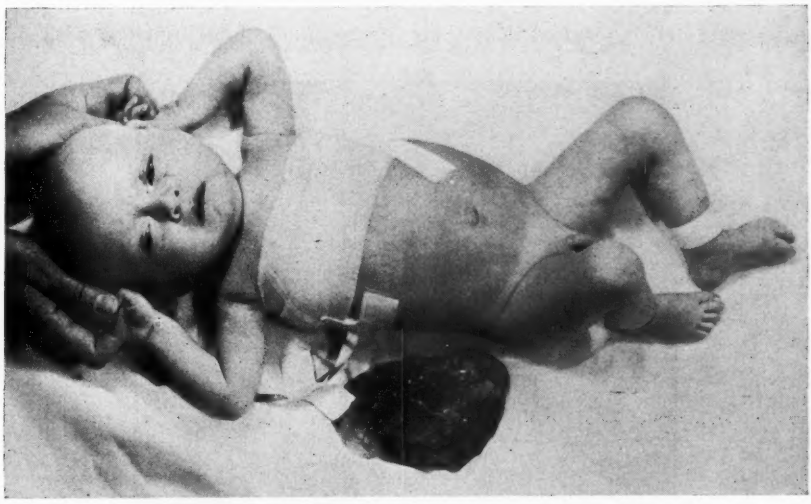

Fig. 3.—Photograph of infant, forty-eight hours after surgery, with the removed middle lobe. Note the size of this lobe compared with that of the baby's chest.

DISCUSSION

This case is significant because it is only the third proved published case of complete absence of the anterior mediastinum undoubtedly of congenital origin. However, of further interest is the possible relationship of the absent anterior mediastinum to the development of the obstructive emphysema. No definite abnormality of the right middle bronchus could be determined either grossly at the time of operation or microscopically in the pathologic specimen. Unfortunately, the condition of the infant prevented bronchoscopy and bronchograms, and an obstruction at the origin of the right middle bronchus may have been overlooked. However, the gradual development of symptoms with the initial symptoms at 6 weeks of age, the presence of the right middle lobe herniated through the wide congenital defect of the mediastinum, and the apparent absence of abnormalities of the bronchus to account for the emphysema

cause one to suspect that the anterior mediastinal defect may not have been purely coincidental, but may have contributed to the development of the emphysema.

The anterior mediastinum represents the midline partition persisting after expansion of the parietal pleurae ceases. Before birth the lungs are relatively small and compact. They occupy only the dorsal portions of the thorax, while the anterior mediastinum is quite extensive and is very broad. With the onset of breathing at birth, the lungs distend with air, assume a more anterior position, and press and narrow the anterior mediastinum. The anterior and central position of the middle lobe make it more vulnerable to herniation through a mediastinal defect. Consequently, in this case we feel that it is highly probable that the herniation of the middle lobe through the anterior mediastinal defect resulted in kinking of the middle lobe bronchus with sub-

Fig. 4.—Anteroposterior roentgenogram on the tenth postoperative day showing the heart in normal position with complete expansion of the right upper and lower lobes.

sequent gradual development of the massive obstructive emphysema. Perhaps one should add this congenital abnormality to the possible etiological agents in obstructive emphysema. The striking lateral films of the chest in this case and the case reported by Gross and Lewis are very suggestive of anterior mediastinal defect, and should justify a presumptive diagnosis preoperatively.

Should such a mediastinal defect be closed? There is insufficient recorded experience to furnish an answer. It would appear that if there were any co-

existent disease or malformation which might unbalance the pressure in the two sides of the chest, the defect should be closed. However, if a defect of the mediastinum were an incidental finding as in Ochsner's case, it would seem better judgment to leave it as found. It is not a simple matter to close a large mediastinal defect. Furthermore, untoward complications conceivably might arise from drawing the pericardial sac with many fine interrupted silk stitches to the periosteum of the posterior surface of the sternum. The mediastinal defect in our patient was not closed because the condition of the patient did not warrant extension of the operating time. It did not seem likely after the emphysematous middle lobe had been removed that there was much chance of further complications from this defect.

SUMMARY

A case report is made of a 4-month-old male infant who had a large anterior mediastinal defect with free interpleural communication and obstructive emphysema of the middle lobe producing severe respiratory symptoms.

The possible role of the anterior mediastinal defect as an etiological agent in the development of the obstructive emphysema in this case is discussed.

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CAVERNOUS HEMANGIOMA OF THE THORACIC WALL ASSOCIATED WITH CLINICAL EVIDENCE OF ARTERIOVENOUS FISTULAS

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WE ARE reporting data on a patient recently observed at the Mayo Clinic who had a large cavernous hemangioma associated with clinical evidence of arteriovenous fistulas involving the seventh, eighth, and ninth intercostal vessels of the left posterior thoracic wall. Resection of the lesion was successfully accomplished.

In a review of the literature in English since 1935, we found no case of cavernous hemangioma associated with clinical evidence of arteriovenous fistula involving the intercostal vessels, in which surgical treatment had been carried out. Cavernous hemangiomas of the thoracic wall which are not associated with clinical evidence of arteriovenous fistula and which require surgical treatment are uncommon enough in themselves. Such lesions involving the bones of the thoracic wall have been reported by Bucy and Capp,[1] Mallory,[2] and Dorner and Marcy.[3] Excision of the involved bone with recovery has been the rule, although some of these patients were successfully treated with roentgen therapy. Shallow and his co-workers,[4] in a review of 335 cases of primary hemangiomatous tumors of skeletal muscle, found thirty-nine involving muscles of the thoracic wall, of which four involved intercostal muscles. Of the entire series, a bruit was heard in 1.8 per cent, and a thrill was noted in 0.6 per cent. At least one of the hemangiomas involving the thoracic wall was associated with both a thrill and a bruit. This was the case of Serra, Lavenia, and Frega,[5] in which the lesion was said to involve the muscles of the right hemithorax. Excision was carried out in this case, with recovery.

Elkin and Cooper[6] reported three large hemangiomas of the thoracic wall, two of which were completely excised. A bruit was not specifically associated in the report with any of these, although in three of the nineteen extensive hemangiomas from all parts of the body with which this report dealt, pulsations were present, and in two a bruit was audible, and was interpreted as evidence of large arteriovenous communications.

During the fifteen-year period from 1935 through 1949, fifteen cases of cavernous hemangioma, in which surgical treatment was carried out, were seen at the clinic. In none of these cases was there clinical evidence of arteriovenous fistulas, as indicated by the findings of a thrill or a bruit, nor was there any roentgenographic evidence of erosion of the ribs. Three of the hemangiomas possibly involved intercostal vessels, but since the lesions were only partially excised, their true extent could not be determined.

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Several cases have been reported in which the presumptive diagnosis of arteriovenous fistula has been made, but definitive treatment was not undertaken. The earliest such case which we could find in the literature in English was that of Langmead,[7] reported in 1912. His patient was a woman aged 55 years with tortuous pulsating vessels over the posterior aspect of the thorax, associated with a thrill and a loud systolic bruit, the pitch of which could be altered by pressure. A rounded circumscribed shadow between the third and fourth ribs posteriorly was seen on the roentgenogram of the thorax. This was thought to be an intercostal aneurysm, but a cirsoid aneurysm was also considered. Pathologic proof was not available.

Hench and Horton[8] described bruits in two female patients, in both of whom a continuous murmur was heard in the seventh interspace on the left, just beneath the lower tip of the scapula, posteriorly. There were no other local signs. Pressure over the bruit did not cause slowing of the heart rate. In one of these patients, roentgenographic evidence of scalloping of the inferior borders of the seventh and eighth ribs on the left was seen. The clinical diagnosis in each case was arteriovenous fistula of the intercostal vessels. No attempt was made to treat the condition.

Maliner[9] reported the case of a boy aged 12 years with an epigastric tumor mass "like a bundle of worms" over which a systolic murmur could be heard, which could be made to disappear with firm pressure. The clinical diagnosis was cavernous hemangioma. There was no roentgenographic evidence of involvement of bone. Surgical treatment was not undertaken.

One case has been encountered at the clinic in the past fifteen years, in which an extracardiac bruit suggestive of an arteriovenous fistula involving vessels of the thoracic wall was heard. The patient was a woman aged 36 years, in whom a to-and-fro machinery murmur was observed over the xiphisternal area, extending over the precordium and left axilla, and posteriorly along the medial border of the left scapula. The results of roentgenographic study were negative, and cardiac catheterization did not reveal any abnormality of the heart or great vessels. There were no associated symptoms, and treatment was not felt to be necessary.

We were able to find two cases of posttraumatic arteriovenous fistula involving vessels of the thoracic wall. Shumacker and Carter[10] reported one among 354 cases of traumatic arteriovenous fistulas and arterial aneurysms. In this case, the fistula was between the internal mammary artery and the innominate vein. More recently, Elkin[11] reported the case of a female physician aged 39 years in whom a traumatic arteriovenous aneurysm of the vessels of the right side of the diaphragm was successfully excised. This patient had previously been considered to have an arteriovenous fistula of the ninth intercostal vessels, of more than twenty years' duration, following repeated thoracenteses for pleural effusion. There was a harsh machinery-like bruit with systolic accentuation in this area, which could not be obliterated by pressure. No erosion of the underlying ribs could be seen on roentgenographic examination.

A congenital arteriovenous fistula involving the left pectoralis major muscle was recently reported to have been successfully resected by Maier and Stout.[12] This lesion occurred in a woman aged 30 years in whom a continuous murmur in the precordial region had led to a previous diagnosis of patent ductus arteriosus. It was not until a mass underlying the left breast became palpable that the diagnosis of arteriovenous fistula of the thoracic wall was made. This mass was slowly expanding, and a continuous bruit with systolic accentuation was heard over it. A thrill which could be diminished by firm pressure was also observed. These authors also mentioned having observed a patient with arteriovenous fistulas, one in the thoracic wall, and the other in the opposite lung, which was giving rise to hemoptysis.

CASE REPORT

A white fireman, aged 40 years, was first seen at the Mayo Clinic on July 10, 1950, complaining of a tumor mass of the posterior left portion of the thorax. The family history was noncontributory. The patient had been well all his life except for a traumatic amputation of the terminal phalanx of the left fifth finger. In February, 1948, while opening a fire hydrant, the patient felt something "give" in the region of the posterior axillary line of the left lateral portion of the thorax. There was pain in the area for two weeks. Treatment was strapping of the thorax. Continuous soreness was felt in this area until three or four months later, when a small mass the size of a half dollar was first noted in the left scapular area. This gradually enlarged and there was some discomfort when the patient lay on this area. Three or four months prior to admission, the patient thought that the enlargement was progressing more rapidly than before, he began to have a sense of "pressure" in the left side of the thorax, and the mass became slightly tender. He had lost no time from work, however. There had been a loss of 12 pounds (about 5.4 kg.) in the past year. Needle aspiration of the tumor had been carried out elsewhere, one week before the patient was seen at the clinic, with temporary reduction in the size of the tumor. The contents were not described. The results of a review of the history by systems were entirely negative.

On physical examination, the patient was found to weigh 145 pounds (about 65.8 kg.) and did not appear to be ill. The pertinent findings were as follows: A soft pulsatile mass, approximately 12 by 6 centimeters in size, was found extending from the inferior tip of the left scapula inferolaterally, with no discoloration of the overlying skin. This mass was not tender, and no thrill was palpable. On auscultation over this mass, a bruit with systolic and diastolic components was heard in some areas. It was best heard in the inferior lateral aspect of the mass. Pressure with the stethoscope over this area did not alter the character of the bruit. Blood pressure was 120/74. The pulse was 76 beats per minute.

The laboratory findings were: hemoglobin 13.2 Gm. per 100 c.c. (88 per cent); leucocytes 6,200 per cubic millimeter; serologic reaction for syphilis negative; sedimentation rate (Westergren) 9 mm. in 1 hour; total proteins 7.7 Gm. per 100 c.c.; normal albumin-globulin ratio; calcium 10.6 mg. per 100 c.c. of serum; phosphate 3.0 mg. of phosphorus per 100 c.c. of serum; Bodansky alkaline phosphatase was 2.8 units. The urine was negative except for rare erythrocytes in the sediment.

A roentgenogram of the thorax showed a soft-tissue tumor in the left side of the back, with scalloping defects involving the inferior margin of the seventh rib, both margins of the eighth rib, and the superior margin of the ninth rib (Fig. 1).

The clinical diagnosis was arteriovenous fistula and aneurysm of the posterior thoracic wall. Resection was advised in spite of the probable difficulty and danger involved, because it was felt that there was no alternative to surgical treatment. Accordingly, operation was carried out on July 14, 1950, by one of us (Clagett).

Fig. 1.—Portion of the preoperative roentgenogram of the thorax showing the diffuse haziness of the soft-tissue mass in the posterior wall of the left side of the thorax, with scalloping defects involving the inferior margin of the seventh rib, both margins of the eighth rib, and the superior margin of the ninth rib.

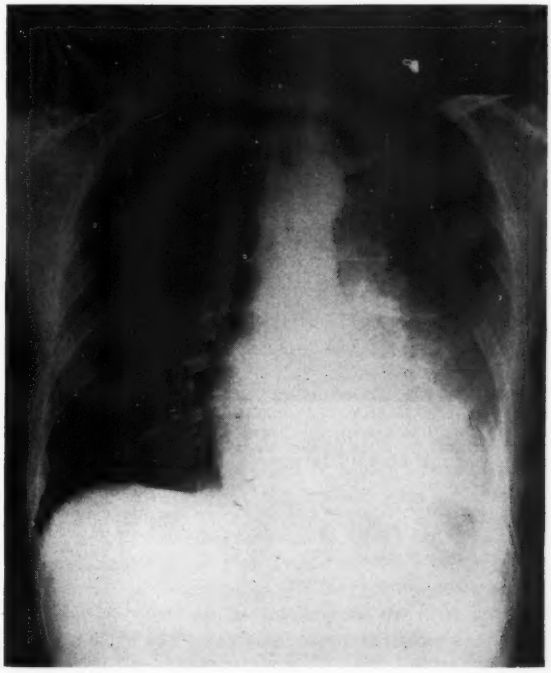

Fig. 2.—Postoperative roentgenogram of the thorax, to demonstrate the size of the defect in the thoracic cage following removal of portions of three ribs which were involved in the cavernous hemangioma.

Operation.—A curved incision was made over the large pulsating mass involving the left side of the thorax posteriorly in the region of the seventh, eighth, and ninth ribs, just below the tip of the scapula. The incision was deepened through the trapezius and latissimus dorsi muscles. The superficial layers showed some evidence of increased blood supply. The large pulsating tumor mass was exposed. It apparently involved the seventh, eighth, and ninth ribs and intercostal spaces and thoracic wall posteriorly. In order to ascertain the intrathoracic involvement, the ninth rib was sectioned anterior to the mass, and the pleura was opened. Huge tortuous dilated blood vessels involving a plaque of thoracic wall could be felt. The eighth and seventh ribs were sectioned anterior to the aneurysmal mass to find and ligate the intercostal vessels accompanying these three ribs, and that portion of the thoracic wall was then resected upward. The interior of the pleural space could then be exposed. With considerable difficulty, the seventh, eighth, and ninth intercostal arteries and veins adjacent

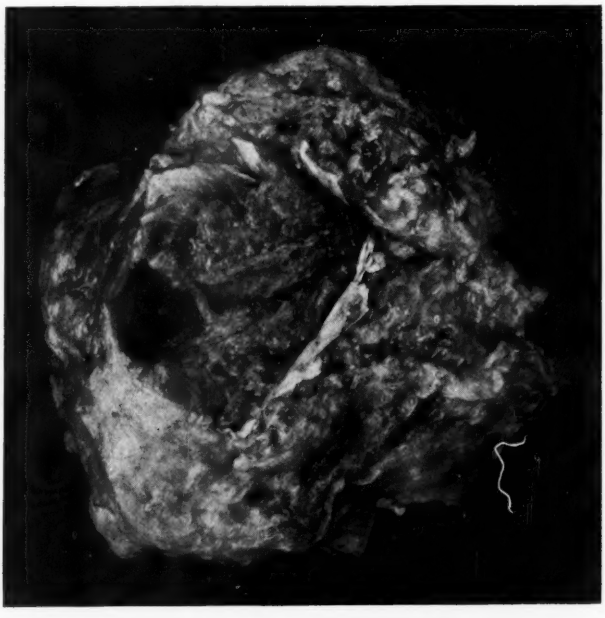

Fig. 3.—Gross pathologic specimen, consisting of a portion of the thoracic wall, showing the musculature overlying the portions of the resected ribs. Both the cavernous hemangiomatous musculature and ribs are involved in the mass.

to the aorta were dissected out, ligated, and divided. The whole tumor mass became softer after this, and resection of the plaque of involved thoracic wall was begun. In spite of the fact that it was thought that the entire blood supply to the thoracic wall had been controlled, on all sides, there was a very severe hemorrhage posteriorly as the aneurysmal mass was cut away. It came directly from the bone marrow of the three ribs where they had been cut posteriorly. This had to be controlled with bone wax. The whole area oozed and bled, but the hemorrhage was finally controlled with mattress sutures and gelfoam. The pleural space was washed with saline solution, and the lung was inflated. A catheter was inserted for temporary closed drainage of the pleural space. Since the defect in the thoracic wall was too large to permit closure, no attempt was made to close the opening between the ribs; however, the trapezius and latissimus dorsi muscles superficially were closed satisfactorily. An

airtight closure of the thoracic wall was made. Although the patient had lost a considerable quantity of blood during the operation, with adequate transfusion (6 liters of whole blood), he remained in good condition.

The postoperative course was uneventful. The catheter in the pleural space was removed on the third postoperative day, and the patient was dismissed from the hospital on the tenth day. The concentration of hemoglobin following operation was 13.2 Gm. per 100 c.c. Fig. 2 shows the size of the defect in the thoracic cage produced by the operation.

Pathologic Findings.—Gross: The specimen consisted of a portion of the thoracic wall measuring 12 by 7 by 6 cm., including pleura, a portion of three ribs, and intervening intercostal muscles, and adjacent musculature of the thoracic wall (Fig. 3). Beneath the smooth pleural surface, dilated blood vessels were visible. When the pleura was peeled away, large tortuous vessels were found lying next to the inferior surface of the seventh rib, the superior and inferior surfaces of the eighth rib, and the superior surface of the ninth rib, all of which were markedly eroded. These vessels communicated directly with large, thin-walled sinuses in the intercostal muscle bundles, and with a spongelike mass of smaller sinuses lying just outside the ribs in the adjacent musculature. The intercostal muscles were partially replaced by the blood vessels and sinuses. A vessel approximately 3 mm. in diameter entered the superior surface of the eighth rib directly, through a canal perforating the cortex of the rib. The marrow cavities of all three ribs were unusually vascular in appearance.

Microscopic: There were many large irregular, and innumerable small endothelial-lined spaces involving striated muscle, areolar tissue, and bone. The wall of some of the larger vascular spaces contained no smooth muscle; in others, a thin, poorly defined smooth muscle layer was present, and in still other larger spaces, a well-defined medial layer of smooth muscle, and an internal elastic lamina could be seen. The endothelium was heaped up in some of the spaces. In addition to the larger vascular spaces, there were sheets of proliferated endothelial cells, some areas of which suggested capillary formation. The cortices of the ribs contained numerous large vascular spaces, communicating with normal-appearing marrow.

COMMENT

Extracardiac vascular bruits of the thorax can be heard in cases of pulmonary arteriovenous fistula, and of fistulas and aneurysms involving the intrathoracic great vessels. The diagnosis in these instances is usually strongly suggested by the clinical and roentgenographic picture. Cavernous hemangiomas of the breast (Sherry),[13] muscle (Shallow and co-workers)[4] and liver (Lichtman)[14] are also said to give rise to bruits, hums, or murmurs, occasionally. This would support the contention of Reid,[15] which seems to be gaining general acceptance, that cavernous hemangiomas have the same etiologic basis as congenital arteriovenous fistulas and cirsoid aneurysms. All are believed to result from a developmental error in which the capillary bed normally intervening between the arterial and the venous side of the vascular tree is lacking, and abnormal arteriovenous communications are present, or potentially present. As Lewis[16] has pointed out, the actual communication or communications between artery and vein may not be established until relatively late in life, and then may occur either spontaneously or following some injury. This would help to explain why symptoms and signs of these supposedly congenital lesions may not be noted in many instances until adulthood. Our case is an illustration of such a probability, in which the presence of a congenital lesion was not apparent until a muscular injury of later life resulted in large enough arteriovenous communications to produce the symptoms and signs described.

Arteriovenous fistulas involving intercostal vessels are rare. Not a single case could be found among 447 cases of arteriovenous fistula reported in Callander's[17] series, nor among the 620 cases, the data on which were analyzed by Matas[18] from his own personal experience.

Notching or scalloping of ribs is usually associated with coarctation of the aorta. However, Batchelder and Williams[19] have described notching of the inferior surfaces of the fifth, sixth, and seventh ribs on the left, posteriorly, in the case of a man aged 38 years with the tetralogy of Fallot, in which at necropsy, dilated, tortuous intercostal arteries were found in these locations, without coarctation of the aorta or aneurysm of these vessels. Maier and Stout[12] also mentioned notching of the ribs, in a case of arteriovenous fistula of the thoracic wall in association with a pulmonary arteriovenous fistula, to which we have already referred. Maier mentioned this observation again, in discussing the report of Lindskog and his co-workers,[20] on pulmonary arteriovenous aneurysms. Our case illustrates this unusual cause for scalloping of the ribs yet again.

SUMMARY

A case has been reported of cavernous and capillary hemangioma of the left thoracic wall, with clinical evidence of arteriovenous fistulas, in which resection of the involved ribs, intercostal muscle bundles, and adjacent musculature of the thoracic wall was successfully carried out. The pertinent available literature is reviewed, and the significance of extracardiac bruits and of notching of the ribs, both of which were present in this case, is discussed.

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AN ANOMALY OF THE PULMONARY VEINS

A Case Study

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CONGENITAL malformations within the pulmonary circulation form a very interesting group of vascular anomalies. Once thought to be extremely rare and diagnosed only at autopsy, these lesions are being encountered more and more frequently by both clinician and surgeon. The increased use of exploratory thoracotomy, angiocardiography, and cardiac catheterization as diagnostic aids will no doubt bring many more of these cases to our attention. In the case reported here the right inferior pulmonary vein drained into the inferior vena cava. There were associated malformations of the lobar bronchi and an aberrant bronchial artery to the right lower lobe. The anomalies were discovered at operation and further evaluated by angiocardiography, cardiac catheterization, and bronchospirometry.

A 20-year-old, white male soldier was admitted to a station hospital in Japan because of a persistent shadow on roentgenogram examination which suggested segmental atelectasis in the right lower lobe. The patient had successfully undergone basic training and endured the rigorous physical exertions required of an infantryman without symptoms. He was transferred to Walter Reed Army Hospital for diagnosis and disposition. Roentgenograms revealed a crescent-shaped area of increased density in the right lower lung field, which paralleled the right heart border (Fig. 1).

Physical examination was negative. There was no history of cyanosis or dyspnea. The patient had developed normally in childhood, and his activities had in no way been curtailed. Pulmonary function studies were within normal limits. Bronchoscopy revealed some variation in the right lobar bronchi in that the lower lobe bronchus divided into two equal sized bronchi from which the segmental bronchi arose. One of the segmental bronchi seemed stenosed or occluded. The remainder of the bronchoscopic examination was negative. Bronchography substantiated the bronchoscopic findings (Fig. 2).

The more lateral of the two lower lobe bronchi gave off segments which seemed to correspond to the superior segmental bronchus of the lower lobe, the two segmental bronchi of the middle lobe, and the anterior basal and lateral basal bronchi of the lower lobe. The medial of the two main lower lobe bronchi gave off two segments which corresponded to the posterior basal and medial basal segmental bronchi. In addition, there were two segmental bronchi which appeared to be blocked or stenosed. It was on the basis of this evidence that the diagnosis of segmental atelectasis was made, and exploratory thoracotomy advised.

At operation, a few filmy adhesions were present between the lower lobe and diaphragm. As these were stripped away a large vein was visualized on the diaphragmatic surface of the lower lobe. The vein appeared to take origin from within the lung structure and pass through the central tendon of the diaphragm (Fig. 3).

Accompanying the vein was a large artery which entered the under surface of the lobe from below the diaphragm. Further exploration revealed complete absence of the pulmonary ligament and no inferior pulmonary vein in the region where it is ordinarily located. The superior pulmonary vein was easily visualized, and appeared normal. The pulmonary artery also appeared normal. In the region where bronchography led us to sus-

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pect stenosed or occluded bronchial segments, blind pouches from otherwise normal bronchi could be palpated. The anomalous vein which seemed to provide the only drainage for the right lower lobe was apparently the inferior pulmonary vein. As the exposed anomalous vein, which was approximately three centimeters in length, approached the diaphragm it was directly in line to join the inferior vena cava, which could be easily visualized because of the absence of the pulmonary ligament. The two veins united just

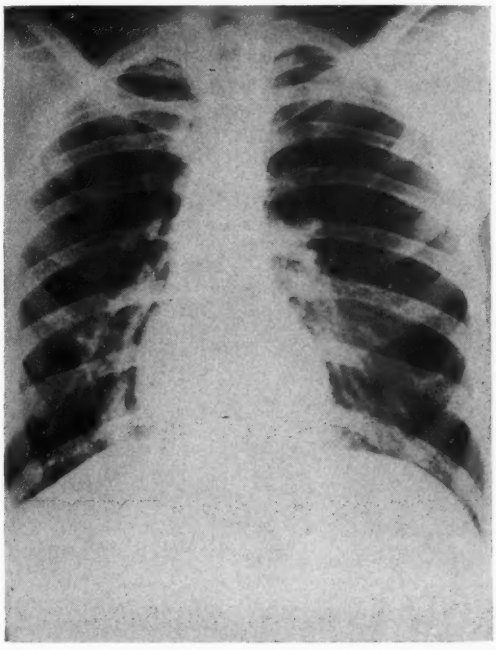


Fig. 1.—Posterior-anterior roentgenogram of the chest.

beneath the diaphragm. Exploration of the right hemithorax revealed no other abnormalities. The heart appeared normal except for prominence of the right border. On the basis of these findings, it was decided that further studies which should include angiocardiograms and cardiac catheterization would be necessary to determine whether or not other abnormalities existed. The chest was closed. The patient had an uneventful convalescence.

Angiocardiograms were done six weks after the exploratory thoracotomy. A series of eight angiocardiograms was made at one and one-half second intervals following diodrast injection (Figs. 4*A* and 5*A*). There were no abnormalities of the cardiac chambers, pulmonary arteries, or aorta. The left auricle was well visualized, and at the time of opacification of this chamber and the pulmonary veins, an unusual, irregular prominent network of vessels appeared in the right middle lung field continuous with a large vessel along the right cardiac border. The latter was approximately 18 millimeters in diameter and paralleled the curve of the right cardiac border from which it was separated by seven millimeters. This vessel seemed to pass beneath the diaphragm.

The filling of this vessel at this particular time leaves no doubt that it was a large pulmonary vein connected with an abnormally prominent and irregular network of pulmonary veins in the right middle lung field. No arteriovenous fistulae were demonstrated.

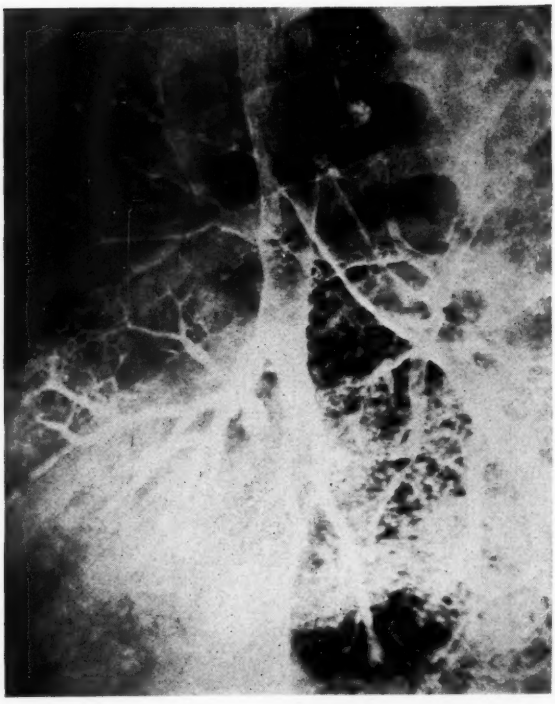


Fig. 2.—Bronchography—left anterior oblique.

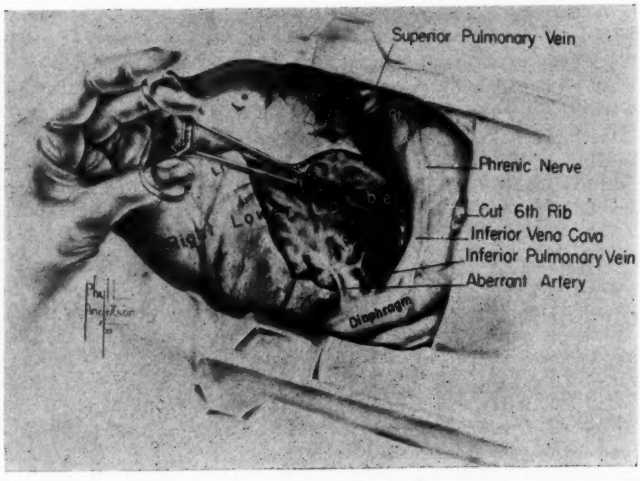
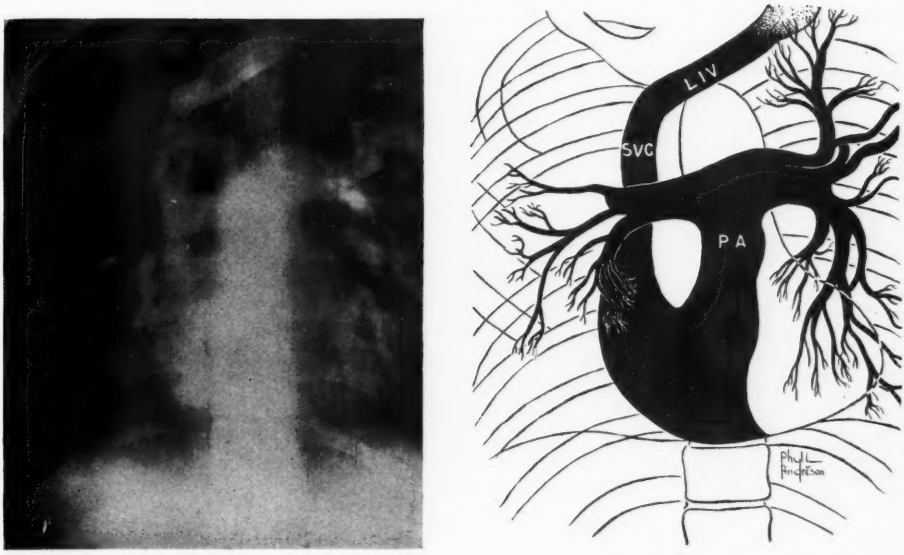
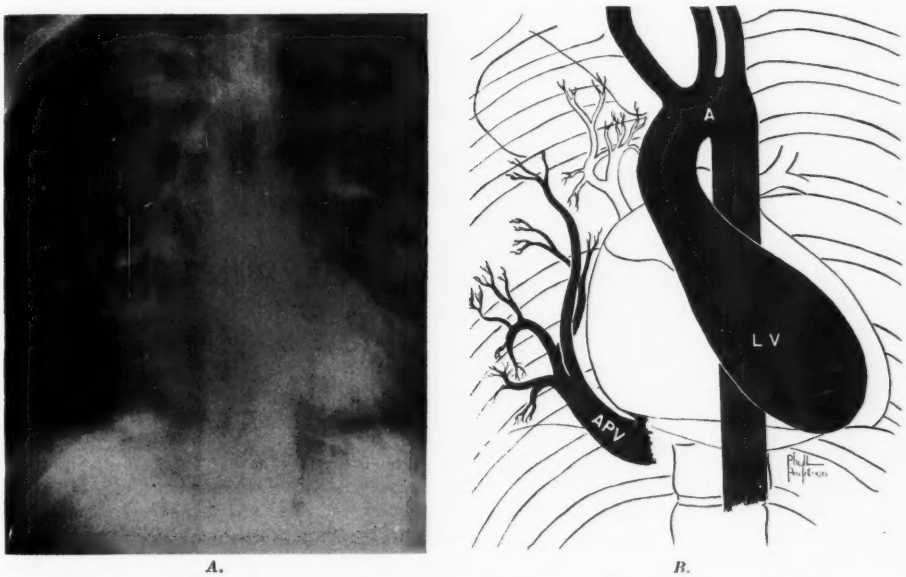


Fig. 3.—Artist's conception of the anomaly as seen at operation.



A. *B.*

Fig. 4.—Angiocardiogram three seconds after intravenous injection of 70 per cent diodrast showing opacification of superior vena cava, the right side of the heart, and the pulmonary arteries. *LIV*, left innominate vein. *PA*, main stem of pulmonary artery. *SVC* superior vena cava.



A. *B.*

Fig. 5.—Angiocardiogram seven and one-half seconds after intravenous injection of seventy per cent diodrast. The anomalous pulmonary vein becomes opacified at the same time as the left side of the heart and aorta. *A*, aorta. *APV*, anomalous pulmonary vein. *LV*, left ventricle.

The cardiac catheterization was performed twelve weeks postoperatively, at which time the vital capacity had returned to normal. Under fluoroscopic guidance, a catheter was directed into the right heart where the tip was passed into the inferior vena cava with difficulty. After considerable manipulation, the catheter was inserted into the anomalous pulmonary vein (Fig. 6), where it passed laterally and cephalad outside the cardiac shadow to reach the lung hilum. The tip was then withdrawn six centimeters to the midpoint of the vein. The catheter tip was then placed in the inferior vena cava and withdrawn into the right auricle. Samples and pressures were recorded at all locations, and at a final position in the superior vena cava. Since the fluoroscopic time had expired, the catheter was withdrawn without exploration of the right ventricle and pulmonary artery. At a repeat catheterization, attempts to enter the ventricle and pulmonary artery were unsuccessful. These data are tabulated in Table I. Oxygen content of the blood was determined by the method of Van Slyke and Neill.

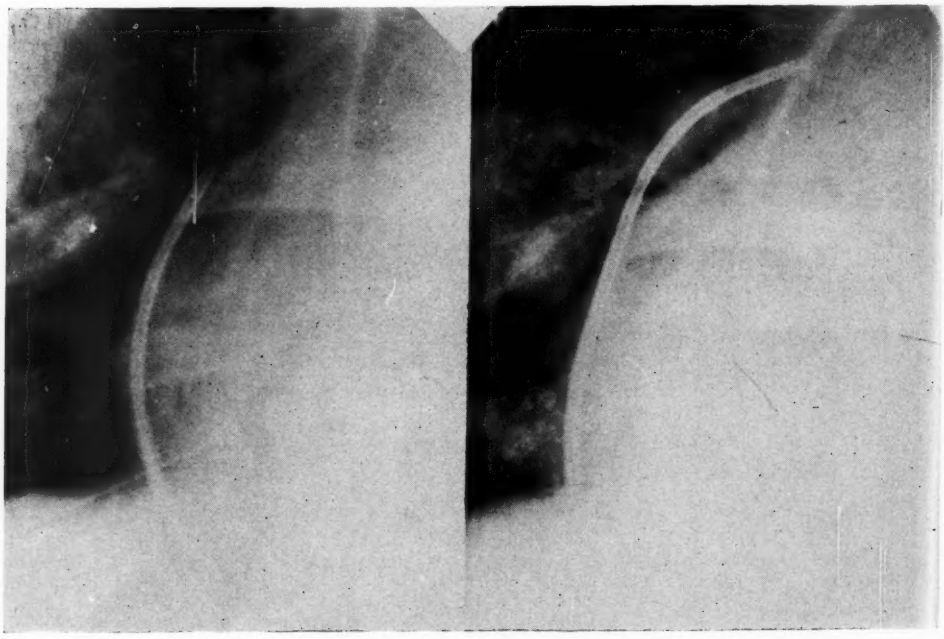


Fig. 6.—Catheter in the anomalous vein.

Blood pressure was recorded by means of an electromanometer. The mean pressure in millimeters of mercury varied from four to six in the pulmonary vein, and from two to six in the right auricle.

From this data we have computed the systemic cardiac output using the average oxygen content of the samples of blood obtained from the superior and inferior vena cava. It is acknowledged that this value does not accurately represent the mixed venous blood and that the output is at best an approximation. The Fick principle is applied with the results shown in Table II.

The output of the right ventricle is greater than that of the left ventricle by the volume of the shunt. Since the venous drainage of the lungs is shared by the four pulmonary veins, it is estimated that approximately one-fourth of the pulmonary venous return enters the right auricle, assuming that flow into the right lower lobe is unimpaired.

TABLE I

	OXYGEN CONTENT VOLS. PER CENT	OXYGEN SATURATION*
Superior vena cava	16.3	73
Inferior vena cava	17.7	80
Right auricle, upper	17.9	81
Right auricle, lower	18.7	84
Pulmonary vein, middle	19.0	86
Pulmonary vein, near hilum	21.5	98
Femoral artery	21.8	98

*Average oxygen capacity—22.2 volumes per cent.

TABLE II

Oxygen consumption ml./min.	280*
Oxygen content Mixed venous blood ml./liter	170
Oxygen content Arterial blood ml./liter	218
Oxygen arteriovenous difference m/liter	48
Left ventricular output l./min.	5.8*

*These figures represent resting but not basal conditions.

On this basis, the right ventricular output will be approximately one-third or 1.9 liters greater than the left ventricular output.

Bronchospirometry was done, and demonstrated the oxygen uptake of the right lung to be 48.8 per cent of the total oxygen consumption, and the vital capacity of the right lung was 44.6 per cent of the total. The normal oxygen consumption of the right lung and the normal oxygen saturation of the blood obtained from the anomalous vein would seem to indicate that the right lower lobe is ventilating and respiring normally.

Blood volume determinations by use of the Evans Blue method and also by untilizing radioactive phosphorus in each instance were below the expected average normal.

TABLE III

Total blood volume (radioactive phosphorus)	3,700 c.c.
Total blood volume (Evans Blue method)	4,100 c.c.
Estimated normal for weight	5,200 c.c.

DISCUSSION

We are indebted to A. J. Brown[1] for much of our knowledge of the development of the pulmonary veins. His very thorough observation was made using embryos of the domestic cat. According to Brown,[1] the primitive pulmonary vein is formed from the capillary plexus that surrounds the foregut, and is distinctly visible in embryos as small as 5 mm. Later observers[2] have described this vascular channel in 2.5 mm. embryos. The division of the tracheal bud from the foregut is completed by the fourth week.[3] As the tracheal bud continues to develop in a ventral and caudal direction, it carries with it a portion of the large plexus of vessels which form a network about the entire length of the gut tube. These vascular channels designated by Brown as the splanchnic plexus communicate freely with both the precardinal and postcardinal venous systems. In the cephalic portion of the splanchnic plexus which is in the region of the protruding lung bud, two main vascular channels appear. These fuse in the midline to form a single vessel which runs caudad and ventrad piercing the dorsal mesocardium and entering the sinus venosus

in the midline. This is the primitive pulmonary vein. Numerous anastomoses are apparent between the primitive pulmonary vein and the splanchnic plexus on the foregut. In the caudal part of the splanchnic plexus in the region of the developing liver, another large vascular channel is formed as a result of the fusion of the capillaries of the splanchnic plexus in this region. This vessel runs cephalad and ventrad entering the sinus venosus, and represents the primitive posthepatic portion of the inferior vena cava. This primitive cava has numerous anastomoses with the splanchnic plexus including that portion which is carried forward by the developing lung and is at this time recognized as the pulmonary plexus. The malformation found in this case is a result of a persistence of one of the vascular channels which in the embryo represents a normal anastomosis between the pulmonary plexus and the primitive posthepatic vena cava. The persistence of this channel may have been due to the failure of development of adequate drainage of the lower lobe, or perhaps its persistence made unnecessary the formation of a normally situated inferior pulmonary vein so that the anlage of that structure disappeared early in embryonic life.

A case of this type reported by Park[4] in 1912 was quoted by Brown to substantiate his observations on the development of the pulmonary veins. The case reported by Park, however, had other cardiac abnormalities which caused death at the age of two and one-half years.

Brodie[5] in 1942 summarized the 106 cases reported up to that time in which blood returning from the lungs re-entered the right heart; of these 68 were cases of incomplete pulmonary venous drainage into the right heart similar to the case reported here. In a later review, Edwards and Dushane[6] found a total of 132 cases of anomalous pulmonary venous drainage established by autopsy. The anomaly has recently been demonstrated during life by angiocardiography and cardiac catheterization,[7-11] and at operation by Brantigan[12] and Drake.[13]

The clinical significance of this anomaly and its therapy are not well understood. It is apparent from these studies that there exists in this type of case a shunt of arterial blood back into the venous circulation. It is estimated that one-fourth of the oxygenated blood returning from the lungs is reentering the right side of the heart, and only three-fourths of the blood returning from the lungs is available for transport of oxygen to the tissues. Since sufficient oxygen is available for long and sustained physical effort, as has been repeatedly demonstrated in this patient, it is obvious then that the right ventricle is supplying the necessary amount of oxygenated blood to the left ventricle despite the loss of 25 per cent of this blood in transit. The stroke volume of the right ventricle is one-third greater than that of the left ventricle, therefore we presume that the right ventricle has a larger capacity than the left. This supposition is borne out by clinical observations on the prominence of the right heart border in roentgenograms and also at operation. The normal electrocardiograms would suggest that the increased right ventricular output is well compensated and not associated with myocardial hypertrophy or strain.

The pulmonary function studies are worthy of comment. Dotter[9] and associates, on the basis of their observations of two patients in whom the entire right pulmonary venous drainage is into the right auricle, have suggested that there may be diminished respiratory function of the abnormally drained lung. Observations in this case have uncovered no evidence to support this opinion, but, on the contrary, the involved lung tissue in this case appears to be functioning in a normal manner.

SUMMARY

A study is reported of a patient in whom the right inferior pulmonary vein drained into the inferior vena cava. The anomaly was observed at surgery and studied by angiocardiography, cardiac catheterization, and bronchospirometry.

The function of the involved lung appears normal. The output of the right ventricle is increased, but there is no evidence at this time of right ventricular hypertrophy or cardiac strain.

Surgical intervention does not seem to be indicated at the present time. The patient was returned to full military duty. Only careful follow-up studies of this case and thorough evaluation of similar cases will indicate whether corrective measures should be employed.

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CONGENITAL ATRESIA OF THE ESOPHAGUS TREATED BY A ONE-STAGE PRIMARY ESOPHAGOGASTROSTOMY EMPLOYING A RIGHT TRANSPLEURAL APPROACH

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CONGENITAL atresia of the esophagus with or without an associated tracheo-esophageal fistula is a serious anomaly, and if not surgically treated it will cause the death of the patient. In about 90 per cent of the cases, the proximal segment of the esophagus ends in a cul-de-sac, and the distal segment is connected to the trachea or main stem bronchus by a fistulous communication. Fortunately, in the majority of the patients in this group, closure of the fistula and restoration of esophageal continuity by a primary end-to-end anastomosis may be performed. However, in about 10 per cent of the cases, an associated tracheo-esophageal fistula is not present. In such instances, the distal segment of the esophagus is either entirely absent (agenesis) or so hypoplastic as to preclude a primary anastomosis with the proximal segment of the esophagus.

In the surgical management of patients with congenital atresia of the esophagus, the determination of the presence or absence of an associated tracheo-esophageal fistula is important. In the diagnosis of a fistula between the trachea and the esophagus, Shaw[14] and, more recently, Byron[2] advocated direct endoscopy. Bigger[1] objected to instrumentation of the trachea because of the danger of producing a traumatic subglottic edema which might necessitate an emergency tracheostomy. Instead, Bigger advised the direct instillation of iodized oil into the trachea. The plain roentgenogram of the abdomen has proved a simple but valuable diagnostic aid. In the absence of gas shadows in the stomach or intestines, one may assume that a fistula between the trachea and the lower esophageal segment is absent. However, this may be an erroneous assumption as demonstrated by Haight.[5, 7] This author operated upon seven patients with congenital atresia of the esophagus in whom preoperative roentgenograms of the abdomen did not reveal air shadows in the stomach or intestines. In three of the seven patients, the presence of a tracheo-esophageal fistula was established. In two of these three, a primary anastomosis between the esophageal segments was performed. However, the functional result was not satisfactory in either of these patients because of the extreme diminution in the caliber of the lumen of the lower esophageal segment. Accordingly, one may conclude that in the absence of gas shadows in the stomach or intestines, the actual determination of the presence of a fistula between the trachea and lower segment of the esophagus preoperatively

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would be more of academic rather than practical surgical significance. Under such circumstances, it is believed that an attempt at a primary restoration of esophageal continuity should not be done.

The case herein reported is that of a patient with congenital atresia of the esophagus and a hypoplasia of the lower esophageal segment. An associated tracheo-esophageal fistula was not present. A preoperative roentgenogram of the abdomen showed the absence of gas shadows in the stomach and intestines (Fig. 1, *A*). The surgical treatment employed was a one-stage primary esophagogastrostomy, using a combined extrapleural and intrapleural approach from the right side. To the knowledge of the author, this is the first time that this mode of approach has been reported in the performance of a primary esophagogastrostomy for congenital atresia of the esophagus.

CASE REPORT

The patient, a premature white girl, weighing 5 pounds, was born at 8:30 P.M., Dec. 12, 1949, in St. Clare's Hospital. The delivery was spontaneous and was accomplished without difficulty. When the infant was admitted to the nursery, cyanosis of the upper and lower extremities was noted. The use of a heated crib was prescribed, and oxygen was administered. Routine formula feedings were begun on the day after delivery, but were taken poorly from the start. Attacks of cyanosis with each feeding were observed. The secretion of oronasal mucus was excessive. There was no passage of stool.

Physical Examination: The patient was a premature infant and in generally poor condition. The cry was weak, and a diffuse mottled cyanosis of the skin was present. Salivation was increased. No abnormalities of the heart, lungs, or abdomen were observed. There was a congenital absence of the left thumb and a rudimentary deformity of the right thumb.

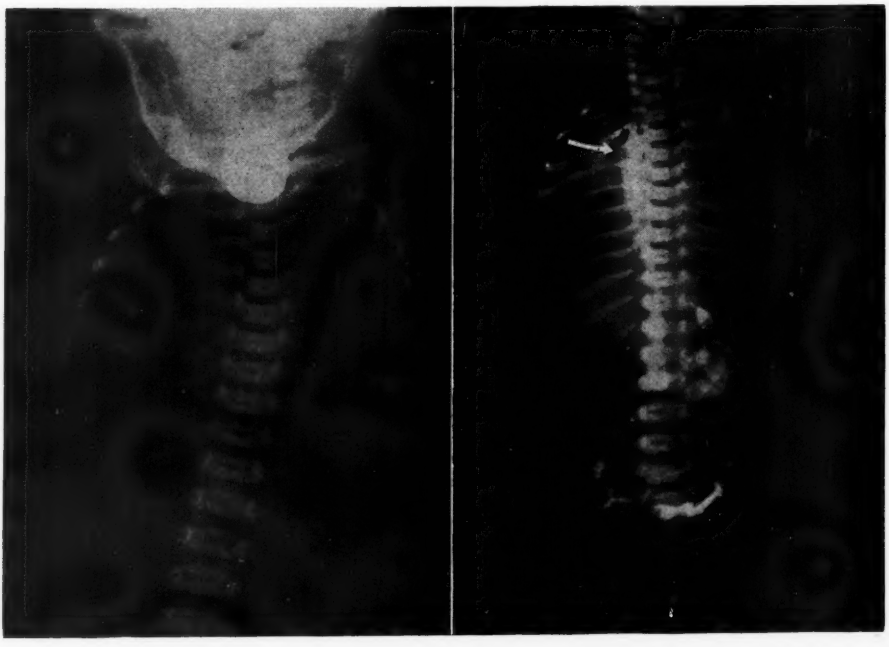
Laboratory Data: The red blood cell count was 3,850,000, and the hemoglobin was 12.5 Gm. (86 per cent). The white blood cell count was 6,100, and the differential cell count was normal. The urinalysis was normal.

Hospital Course: On the second day of life, the patient was seen in consultation by Dr. Charles O'Regan who suggested the diagnosis of congenital atresia of the esophagus. To confirm this diagnosis, a No. 8 French catheter was inserted through the nose into the esophagus. An obstruction to the passage of the catheter was immediately observed. Lipiodol (3 c.c.) was instilled through the catheter, and a roentgenogram was taken (Fig. 1, *A*). This showed that the esophagus ended in a cul-de-sac. A communication with the trachea was not present. The lipiodol was removed by aspiration. A roentgenogram of the abdomen revealed the absence of gas shadows in the gastrointestinal tract. A roentgenogram of the chest showed a fine mottling in both lung fields which was interpreted as a "patchy" atelectasis. A barium enema study showed that the large bowel was patent to the ileocecal region (Fig. 1, *A*).

Following the completion of the diagnostic study, a diagnosis of congenital atresia of the esophagus without tracheo-esophageal fistula was made, and surgical treatment was advised. The patient was prepared for surgery by the administration of 55 c.c. of whole citrated blood, followed by an infusion of 50 c.c. of 5 per cent glucose in distilled water. Penicillin (20,000 units) and streptomycin (125 milligrams) also were administered. On Dec. 15, 1949, three days after birth, operation was performed. Endotracheal ether anesthesia was employed.

Operation: The patient was placed in the direct left lateral prone position, and a curvilinear parascapular incision on the right side was made (Fig. 2, *A*). The incision was deepened through the underlying muscles, and the rib cage was exposed. A superiosteal resection of the fourth rib and a resection of short segments of the third and fifth ribs at

their respective angles was performed. By digital dissection in the extrapleural plane, the parietal pleura was separated from the surrounding ribs, and the azygos vein was exposed. This vein was isolated, doubly clamped, divided, and each end ligated with black silk (00) ligatures. The proximal segment of the esophagus was identified by palpation of the indwelling catheter. This segment was partially mobilized by a combination of blunt and sharp dissection. A fistulous communication with the trachea was not present. In the identification of the lower segment of the esophagus, difficulty was encountered. The trachea, carina, and main stem bronchi were clearly exposed, but there was no communication between these structures and the lower esophageal segment. As dissection was con-



A. *B.*

Fig. 1.—*A*, Preoperative roentgenogram demonstrating the blind upper segment of the esophagus filled with lipiodol, and the absence of gas shadows in the gastrointestinal tract. The barium filled colon is partly visible. *B*, Postmortem roentgenogram demonstrating the site of esophagogastrostomy (arrow) and the presence of barium in the proximal small intestine. The colon is still outlined by residual barium.

tinued in the extrapleural plane, downward toward the diaphragm, a markedly hypoplastic lower segment of the esophagus was seen (Fig. 2, *B*). This finding precluded a primary anastomosis of the esophagus, and an attempt at a primary intrapleural esophagogastrostomy in one stage was believed advisable. Accordingly, the skin incision was extended downward, forward, and medialward to the costal margin. The underlying osseous rib cage was divided in the same plane, and the right pleural cavity was entered. The esophageal hiatus was dilated by digital dissection, and a circular division of the underlying reflected layer of peritoneum was performed. To aid in the mobilization of the stomach, clamp traction was applied to the hypoplastic lower esophageal segment, but in doing so an avulsion laceration of the esophagus at the cardia occurred (Fig. 3, *A*). Resection of this hypoplastic segment was required. Traction sutures of silk were then inserted through the wall of the stomach on either side of the cardia, and mobilization of the stomach into the

right pleural cavity was continued (Fig. 2, *B*). The left gastric vessels were clearly visualized, doubly clamped, divided, and ligated. This permitted the mobilization of the stomach into the right pleural cavity. The cardia was mobilized up to the apex of the pleural cavity, and a two-layer esophagocardiostomy, using interrupted sutures of fine silk (0000) was done (Fig. 4). Despite the extensive mobilization of the stomach, the spleen was not seen. A Levin tube was inserted through the anastomotic stoma, and a rectangular-shaped free graft of parietal pleura was used to enclose the region of the anastomosis (Fig. 4). The right pleural cavity was drained through a water-seal catheter, and the wound was closed in layers with interrupted silk sutures. The duration of the operation was three hours and forty-five minutes. The patient withstood the operation well, and was returned to the ward in satisfactory condition.

Fig. 1*C*.—Comparative roentgenogram of a patient with congenital atresia of the esophagus and tracheoesophageal fistula. The blind upper segment of the esophagus is outlined and gas shadows are distributed throughout the gastrointestinal tract.

Postoperative Course: Immediately following the completion of the operation a roentgenogram of the chest showed a satisfactory expansion of the lungs. The infant was placed in an incubator and oxygen was administered. A transfusion of 75 c.c. of whole citrated blood was given. The temperature was 99.8° F., the respirations were normal, and the color was good. In general, the immediate postoperative condition of the patient was satisfactory. On the first postoperative day a repeat roentgenogram of the chest showed good aeration in both lung fields. A blood count showed 4,150,000 red blood cells, and a hemoglobin value of 12 grams. The white blood cells numbered 8,000, and the differential cell count was normal. A second blood transfusion of 50 c.c. of whole citrated blood was given. A plain film of the abdomen showed the Levin tube in the stomach and gas shadows both in the stomach and the upper portion of the small intestines. During the succeeding part of the day, periodic attacks of cyanosis without any associated respiratory difficulty were observed. In the beginning these attacks were of short duration, but as the day progressed they became more frequent and were prolonged. Repeated examinations disclosed no apparent etiological factor. At 9:05 P.M., Dec. 16, 1949, approx-

imately twenty-six hours following the completion of the operation, the patient became markedly cyanotic and expired. At no time was there evidence of respiratory distress. The necropsy was limited to the reopening of the operative incision and the examination by inspection only of the right pleural cavity and its contents. The right lung was completely expanded, and loosely adherent to the parietal pleura. The esophagogastric anastomosis was intact and it appeared hermetically sealed by the free graft of parietal pleura. There was no obstruction to the passage of a Levin tube. The transplanted portion of the stomach was viable and was not unduly dilated. The cause of death was not determined. However, the heart was not examined and, in the presence of the other associated congenital defects, an abnormality of the heart could not be excluded.

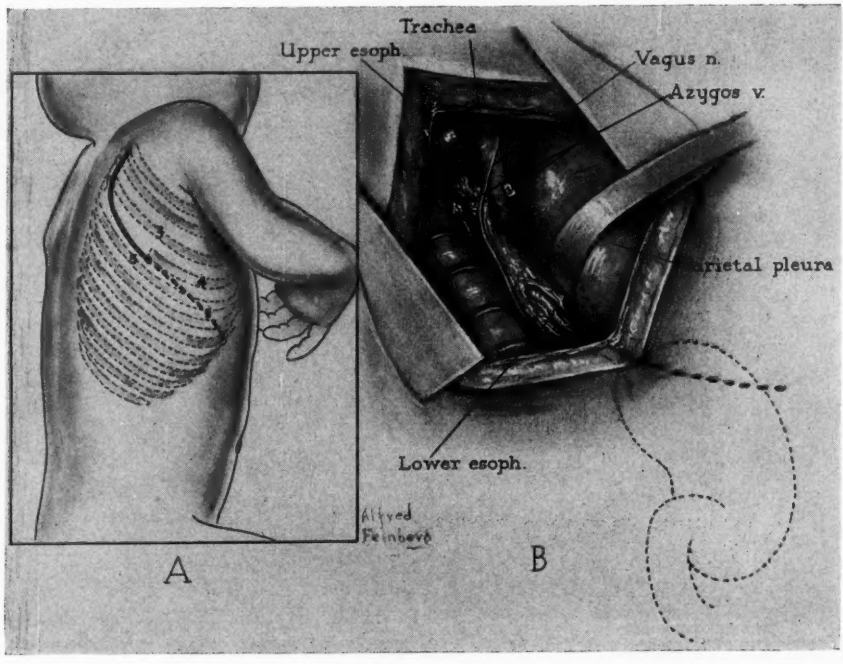


Fig. 2.—*A*, The skin incision is outlined and the extent of resection of the fourth rib is shown. The dotted line indicates the direction of extension of the skin incision. *B*, The extrapleural exposure of the mediastinum from the right side is shown. The dilated proximal segment and the hypoplastic distal segment of the esophagus are demonstrated. The dotted lines indicate the site of incision through the parietal pleura and the line of extension of the skin incision, respectively.

DISCUSSION

In the surgical treatment of congenital atresia of the esophagus with tracheo-esophageal fistula, Lanman,[9] in 1936, was the first to perform a primary anastomosis of the esophagus. An extrapleural approach from the right side was employed with resection of the fourth rib and removal of short segments of the third and fifth ribs. Four patients were treated in this manner, but, unfortunately, all died. Despite this discouragement, Lanman[9] remained of the opinion that this was the operation of choice, and in 1940 predicted that a successful operation would be reported in the near future. In 1939, Shaw[14] per-

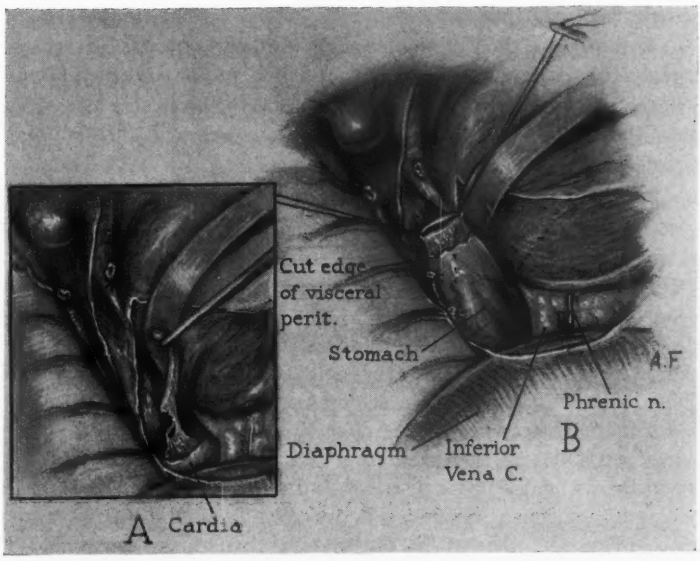


Fig. 3.—*A*, The avulsion laceration of the distal hypoplastic segment of the esophagus at the cardia is demonstrated. *B*, The gastric vessels have been divided and the stomach has been partly mobilized into the right pleural cavity. The dotted line in the proximal segment of the esophagus indicates the site of incision for the anastomosis with the cardia.

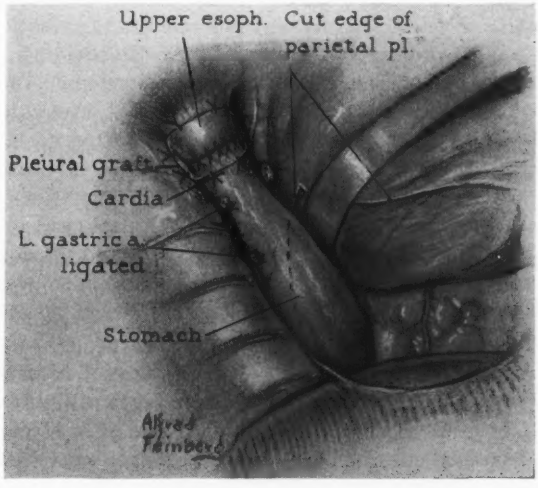


Fig. 4.—The cardia has been mobilized into the apex of the right pleural cavity, the esophagocardiostomy completed, and the anastomosis reinforced by a free graft of parietal pleura.

formed a primary anastomosis of the esophagus using a left paravertebral extrapleural approach and resecting segments of the second to the sixth ribs, inclusive. This patient died on the twelfth postoperative day of a blood transfusion reaction. A necropsy showed separation of the suture line and an infected sinus tract leading to the fistula. Sampson[13] employed a left parascapular incision and resected segments of the third, fourth, and fifth ribs. A primary anastomosis was done, but the outcome was unsuccessful.

In 1941, Haight[4] fulfilled the prediction of Lanman by performing the first successful primary end-to-end anastomosis of the esophagus. An extrapleural approach from the right side was used. The postoperative course of the patient was complicated by leakage at the site of the anastomosis, sinus formation, and subsequent stricture of the esophagus. This responded satisfactorily to bouginage. Following the report of this case in 1943,[4] there was a renewed interest in the surgical management of congenital atresia of the esophagus, both with and without tracheo-esophageal fistula. After an accumulated experience in the treatment of this anomaly, Haight,[5] as did Lanman,[9] recommended an extrapleural approach from the right side, closure of the tracheo-esophageal fistula, and a primary end-to-end anastomosis of the esophagus. The surgical approach from the left side was not favored because of the added technical difficulties inherent in the mobilization of the aorta and its arch. Singleton[15] and, more recently, Lyon and Johnson[12] advocated the transpleural approach and did not consider it an added risk when used in the newborn.

Unfortunately, in about 20 per cent of the patients with congenital atresia of the esophagus, with or without tracheo-esophageal fistula, a primary anastomosis of the esophagus is not possible. Under such circumstances an alternate procedure must be selected. In the past, the multistage operation suggested by Leven[10] (1939) and Ladd[8] (1939), independently of each other, was preferred. This operation consisted of: (1) A preliminary gastrostomy (To obviate leakage at the site of gastrostomy, the Levin tube was inserted through the pylorus into the duodenum). (2) Extrapleural ligation of the fistulous communication between the trachea and the esophagus. (3) Left anterior cervical esophagostomy. (4) Antethoracic esophagoplasty. Although this operation was important as a lifesaving procedure, it was not the ideal in surgical management. Multiple plastic reconstructive operations that required prolonged periods of hospitalization were necessary. Furthermore, the final results were so unsatisfactory that surgeons in general were dissatisfied with this method of treatment.

In 1944, Garlock[3] was the first to report the feasibility of performing a primary esophagogastrostomy, anterior to and above the aortic arch, in the treatment of high-lying lesions of the thoracic portion of the esophagus. In 1946, Swenson[17] successfully utilized this principle in performing for the first time a left transpleural primary esophagogastrostomy in the treatment of congenital atresia of the esophagus with agenesis of the lower esophageal segment. In 1947, Sweet[16, 17] successfully restored continuity of the alimentary canal by performing a secondary intracervical esophagogastrostomy in a patient 21 months of age who, three days after birth, had had a lifesaving multistage

operation performed for congenital atresia of the esophagus with tracheo-esophageal fistula. Subsequently, Byron,[2] then Lyon and Johnson,[12] reported the performance of a left transpleural primary esophagogastrostomy in the treatment of congenital atresia of the esophagus but, unfortunately, in each instance, the patient died. However, these reports confirmed the technical feasibility of the operation and the ability of the young and oftentimes premature infant to withstand the immediate effects of the transpleural approach.

In 1946, Lewis,[11] in the treatment of tumors of the upper and middle third of the thoracic segment of the esophagus, popularized the combined abdominal and right-sided transpleural approach performed in either one or two stages. Recently, the author employed a one-stage right-sided transpleural approach in the successful resection of a carcinoma of the middle third of the esophagus. The esophageal hiatus was enlarged by digital dissection and the underlying layer of peritoneum was divided circumferentially. After ligation and division of the left gastric vessels, the proximal portion of the stomach was drawn into the right pleural cavity. The tumor-bearing segment of the esophagus was widely resected, and an intrathoracic primary esophagogastrostomy was performed. This success suggested the application of this mode of approach in the treatment of congenital atresia of the esophagus. In the past if, on exploration through the conventional right-sided extrapleural approach, the ends of the esophagus were too widely separated to permit a primary anastomosis, recourse was made to a multistage operation with its attendant disadvantages. To avoid this, Singleton[15] attempted to bridge the defect between the segments of the esophagus by incision of the diaphragm through the esophageal hiatus and mobilization of the proximal portion of the stomach into the right pleural cavity. However, the mobilization of the stomach was insufficient because ligation and division of the left gastric vessels were not done. Accordingly, a primary anastomosis of the esophagus was not accomplished.

In the patient herein presented a preliminary roentgenogram of the abdomen showed the absence of gas shadows in the stomach and intestines. This was interpreted as an indication that a primary anastomosis of the esophagus would not be possible. Nevertheless, a right-sided approach was used. An extrapleural exposure of the esophagus disclosed a marked hypoplasia of the lower segment of the esophagus which precluded a primary anastomosis. Rather than perform a multistage operation, the incision was extended across the rib cage, and the right pleural cavity was entered. The esophageal hiatus was dilated by digital dissection and the underlying cuff of peritoneum was divided circumferentially. After ligation and division of the left gastric vessels, the stomach was displaced without difficulty into the right pleural cavity. As the stomach was mobilized it assumed a tubular contour and the cardia, rather than the fundus, remained the uppermost gastric segment. A two-layer anastomosis between the upper segment of the esophagus and the cardia, using interrupted silk sutures, was performed. Although it was unfortunate that the patient died, the ability to perform a one-stage primary esophagogastrostomy through a right transpleural approach was demonstrated.

In the surgical management of congenital atresia of the esophagus, the selection of the mode of surgical approach is determined mainly by the plain roentgenogram of the abdomen. It is assumed that the presence of a fistula between the proximal segment of the esophagus and the trachea has been excluded. In the absence of gas shadows in the gastrointestinal tract a primary anastomosis of the esophagus is not considered. A primary esophagogastrostomy employing the left or right transpleural approach is preferred. In the presence of gas shadows in the gastrointestinal tract, an extrapleural thoracotomy on the right side is employed and, if possible, a primary anastomosis of the esophagus is performed. However, in a certain percentage of the patients in this group, a primary anastomosis of the esophagus may not be possible because of the wide separation of the esophageal segments. If this should obtain, a transpleural extension of the incision is recommended and the stomach mobilized into the right pleural cavity. If, following the mobilization of the stomach, the distal end of the esophagus is viable and the diameter of its lumen is adequate, a direct anastomosis of the esophagus may be performed. Otherwise, a primary intrapleural esophagogastrostomy is preferred. It is believed that every attempt possible should be made to re-establish continuity of the alimentary canal before resorting to a lifesaving multistage procedure.

SUMMARY

A case of congenital atresia of the esophagus with hypoplasia of the lower esophageal segment is presented.

The continuity of the alimentary canal was restored by a primary intrathoracic esophagogastrostomy employing a one-stage combined extrapleural and intrapleural approach from the right side. Death occurred twenty-six hours postoperatively.

CONCLUSIONS

The ideal in the surgical management of congenital atresia of the esophagus is the immediate restoration of continuity of the alimentary canal, either by a direct anastomosis of the esophageal segments or by a one-stage primary esophagogastrostomy.

A primary anastomosis of the esophagus is not attempted if the preoperative roentgenogram of the abdomen shows the absence of gas shadows in the gastrointestinal tract.

A one-stage primary esophagogastrostomy may be performed through a right transpleural approach. Heretofore, the left transpleural approach has been recommended. The approach from the right side permits the mobilization of the stomach through the esophageal hiatus, into the apex of the pleural cavity, and the performance of an intrapleural esophagogastrostomy.

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BENIGN INTRAMURAL TUMORS AND CYSTS OF THE ESOPHAGUS

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TUMORS originating from smooth muscle, connective tissue, nerve sheath elements, and cysts developing from bronchial or esophageal cell rests comprise the majority of surgically significant benign intramural tumors of the esophagus. The fortuitous location of these tumors within the musculature of the esophageal wall with no involvement of the mucosa frequently permits their enucleation without entering the lumen of the esophagus. In a fewer number of cases, esophageal resection will be necessary. Thirty-nine surgically treated benign intramural extramucosal tumors have appeared in the literature. It is the purpose of this paper to record three additional benign intramural lesions of the esophagus—one leiomyoma and two cysts—each successfully treated by enucleation, and to review the literature pertinent to esophageal tumors of this variety.

CASE REPORTS

CASE 1.—J. C. L., N.C.B.H. No. 87275, a 40-year-old man, was admitted complaining of regurgitation of food and a hot burning liquid shortly after meals of fifteen years' duration. On physical examination no pertinent findings were noted. Routine laboratory studies were within normal limits. Barium swallow showed a smoothly outlined filling defect in the wall of the esophagus in its lower third which caused a delay in the passage of the meal and moderate dilatation of the proximal esophagus. Esophagoscopy disclosed a freely movable irregular bulge into the esophageal lumen 40 centimeters from the upper incisor teeth. The mucosa overlying this bulge was intact.

Left thoracotomy was performed through the ninth rib bed. A 7 by 4 by 4 centimeter firm, rubbery, freely movable mass covered by attenuated esophageal muscle presented 4 centimeters above the cardia occupying the left wall of the esophagus. The mass was easily shelled from its intramural bed without opening the mucosa, and the defect in the esophageal musculature was closed with interrupted fine silk. The patient made an uneventful recovery, and was without complaints one year postoperatively. Microscopic examination demonstrated the tumor to be a leiomyoma.

CASE 2.—S. A. D., N.C.B.H. No. 90771, a 39-year-old housewife complained of intermittent dysphagia for two years prior to admission. There had been no regurgitation of food. The physical examination was negative, and routine laboratory studies were within normal limits. Fluoroscopy and roentgenograms of the chest with barium swallow showed a 4 by 6 centimeter mass just below the level of the aortic arch smoothly indenting the esophagus on its right side and posteriorly.

Right thoracotomy was performed through the fifth rib bed. In the posterior mediastinum just beneath the azygos vein, a 5 centimeter cystic mass was noted occupying the muscular wall of the esophagus. The cyst was enucleated from its intramural loca-

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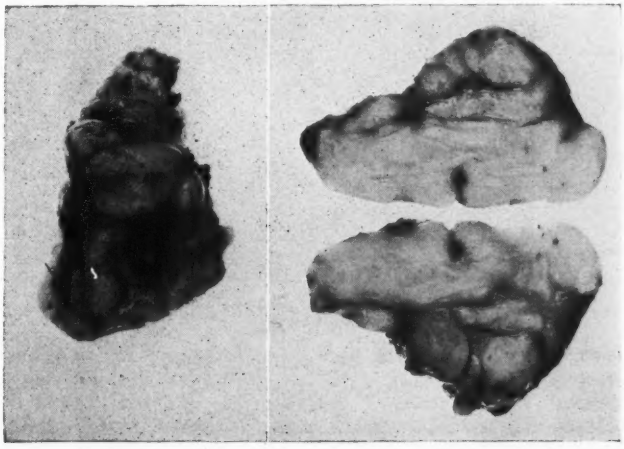
Fig. 1.—Case I. Roentgenogram showing the leiomyoma in the lower third of the esophagus.



A. *B.*

Fig. 2.—*A*, Gross appearance of the tumor from Case 1. *B*, Cut section of the leiomyoma removed in Case 1.

tion without entering the lumen of the esophagus, and the musculature repaired with interrupted fine silk. The patient recovered completely and is well fourteen months following operation. Microscopic examination showed the thin-walled cyst to be lined with transitional type epithelium.

CASE 3.—E. A. S., N.C.B.H. No. 70686, a 53-year-old male complained of a 60-pound weight loss in eighteen months, intermittent almost daily attacks of dull, aching, nonradiation pain in the left lower posterior chest and epigastrium. Three months prior to admission he noted a dry hacking cough which had become progressively worse. There had been no dysphagia. Physical examination was negative. Routine laboratory findings were not remarkable. Chest fluoroscopy with roentgenograms and barium swallow showed in the oblique view a sharply circumscribed tumor mass in the posterior inferior mediastinum which measured 4 by 8 centimeters in diameter.

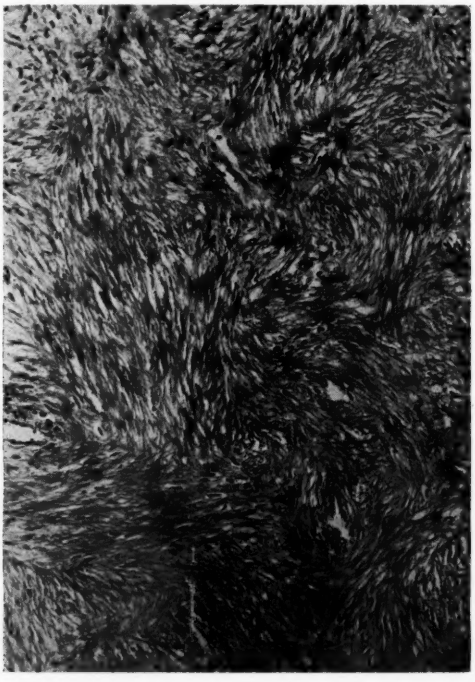


Fig. 3.—Photomicrograph of the tumor removed in Case 1.

The mass was approached from the right side through the bed of the seventh rib. A cystic structure of the dimensions previously noted was found occupying the right wall of the esophagus. It was enucleated from the submucosal structures of the esophageal wall without entering the lumen of the esophagus, although the cyst ruptured in the process with extrusion of a thick gelatinous material. A firm, nonpatent cord one millimeter in diameter connected the esophagus and the cyst at one point. The muscular defect in the esophageal wall was closed with interrupted silk, and the patient's postoperative convalescence was without event. Microscopically the cyst wall contained cartilage and was lined with pseudostratified ciliated columnar epithelium. Follow-up studies showed the patient to be without complaints twenty-seven months postoperatively.

Fig. 4.—Case 2. Roentgenogram showing displacement of the barium-filled esophagus by the cyst which occupied the right lateral wall of the esophagus in the middle third.

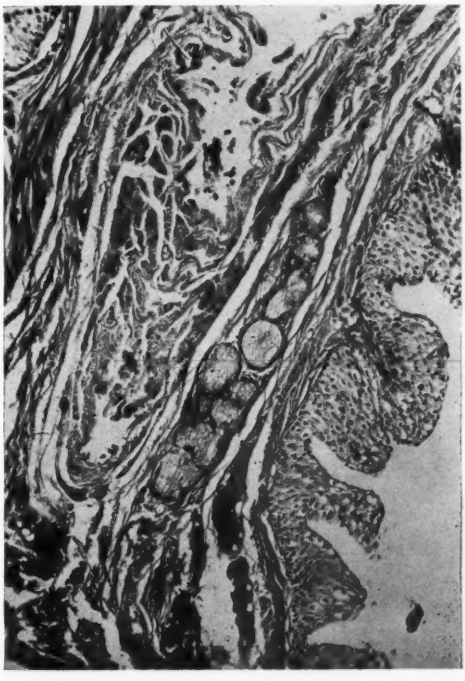


Fig. 5.—Photomicrograph of the cyst wall removed from the patient in Case 2.

Fig. 6.—Roentgenogram of Case 3, showing the posterior mediastinal tumor which proved to be an intramural cyst of the esophagus.

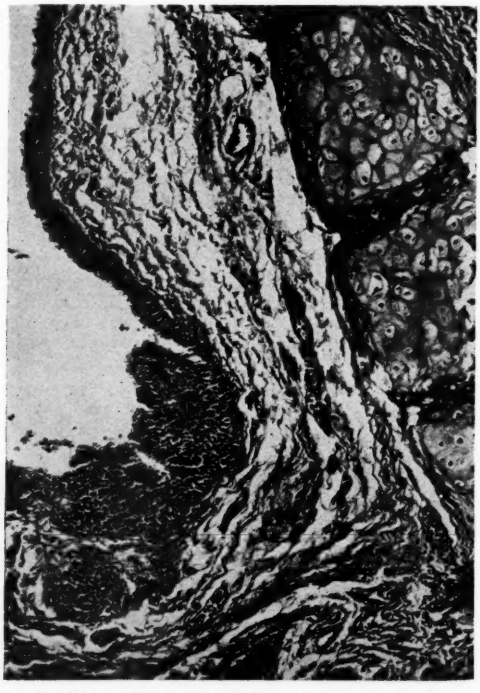


Fig. 7.—Photomicrograph of the specimen from Case 3.

TABLE I. ESOPHAGEAL LEIOMYOMAS REMOVED BY ENUCLEATION

AUTHOR	DATE	AGE	SEX	SYMPTOMS	LOCATION	APPROACH	TREATMENT	DIAGNOSIS	RESULT
Ohsawa[29]	1933	43	F	Dysphagia	Lower third	Transpleural	Enucleation	Myoma	Recovery
Harrington[21] Moersch	1944	56	F	Substernal pain	Lower third	Abdominal	Enucleation	Leiomyoma	Recovery
Harper[19] Tiscenco	1945	41	M	Epigastric distress; vomiting	Middle third	Transpleural	Enucleation	Leiomyoma	Not given
Calmenson[7] Clagett	1946	52	M	Dysphagia; substernal distress	Upper third; middle third (two tumors)	Transpleural	Enucleation	Leiomyomas	Recovery
Nissen[28]	1949	56	M	Dysphagia	Middle third	Transpleural	Enucleation	Leiomyoma	Recovery
Hurwitz[22]	1949	31	M	None	Middle third	Transpleural	Enucleation	Leiomyoma	Recovery
Harrington[20]	1949	31	M	None	Middle third	Transpleural	Enucleation	Leiomyoma	Recovery
Harrington[20]	1949	58	F	Epigastric distress; regurgitation	Lower third	Transpleural	Enucleation	Leiomyoma	Recovery
Goldman[18] Masters	1950	63	F	Dysphagia	Lower third	Transpleural	Enucleation	Leiomyoma	Recovery
Daniel[14] Williams	1950	38	F	None	Lower third	Transpleural	Enucleation	Leiomyoma	Recovery
Bigger[3] Kay Vinson	1950	24	M	Dysphagia	Middle third	Transpleural	Enucleation	Leiomyoma	Recovery
Madden[26] Olmstead	1950	28	M	Cough; dyspnea; chest pain	Upper third	Transpleural	Enucleation	Leiomyoma	Recovery
Dugan[15] Meagher	1950	27	F	Dysphagia; epigastric pain	Middle third	Transpleural	Enucleation	Leiomyoma	Recovery
Cornell[12] Shehadi Sharnoff	1950	54	M	Indigestion	Lower third	Transpleural	Enucleation	Leiomyoma	Recovery
Garlick[16] Stegmaier	1951	48	M	Substernal pain	Middle third	Transpleural	Enucleation	Leiomyoma	Recovery
Myers Bradshaw		40	M	Regurgitation	Lower third	Transpleural	Enucleation	Leiomyoma	Recovery

TABLE II. ESOPHAGEAL CYSTS REMOVED BY ENUCLEATION

AUTHOR	DATE	AGE	SEX	SYMPTOMS	LOCATION	APPROACH	TREATMENT	DIAGNOSIS	RESULT
Sauerbruch[31]	1937	13	M	Dysphagia	Lower third	Transpleural	Enucleation	Embryonic epithelial cyst	Recovery
Churchill[10]	1940	42	M	Cough; substernal pain	Middle third	Transpleural	Enucleation	Bronchogenic cyst	Recovery
Shatzki[33] Hawes	1942	26	F	Regurgitation	Middle third	Transpleural	Enucleation	Bronchogenic cyst	Recovery
Brown[6] Robbins	1944	25	F	Dysphagia; vomiting	Middle third	Transpleural	Enucleation	Bronchogenic cyst	Recovery
Harrington[21] Moersch	1944	39	F	Cough; dyspnea; substernal pain	Middle third	Transpleural	Enucleation	Infected cyst	Recovery
Sweet[34]	1947	43	M	Dysphagia; substernal pain	Lower third	Transpleural	Enucleation	Bronchogenic cyst	Recovery
Bradford[4] Mahon Grow	1947	22	M	Regurgitation	Lower third	Transpleural	Enucleation	Bronchogenic cyst	Recovery
Cornell[11] Blumberg Sarot	1950	1½	M	Cough	Upper third	Transpleural	Enucleation	Bronchogenic cyst	Recovery
Gledhill[17] Morrow	1950	22	F	Dysphagia; substernal pain	Middle third	Transpleural	Enucleation	Epithelial cyst	Recovery
Myers Bradshaw		39	F	Dysphagia	Middle third	Transpleural	Enucleation	Embryonic epithelial cyst	Recovery
Myers Bradshaw		53	M	Chest pain epigastric pain	Lower third	Transpleural	Enucleation	Bronchogenic cyst	Recovery

TABLE III. BENIGN ESOPHAGEAL TUMORS RESECTED OR EXCISED

AUTHOR	DATE	AGE	SEX	SYMPTOMS	LOCATION	APPROACH	TREATMENT	DIAGNOSIS	RESULT
Sauerbruch[31]	1937	33	M	Epigastric distress	Lower third	Transpleural	Resection esophagogastrostomy	Fibromyoma	Recovery
Kornblum[24] Bradshaw	1941	14	F	Dysphagia; vomiting	Lower third	Transpleural	Excision Thorek	Neurofibroma	Died
Brock[5]	1943	47	M	Indigestion	Lower third	Thoracoabdominal	Resection esophagogastrostomy	Myoma	Recovery
Harrington[21] Moersch	1944	39	M	Asthma	Lower third	Transpleural	Resection esophagogastrostomy	Leiomyoma	Died
Schafer[32] Kittle	1947	46	M	Epigastric distress; back pain	Lower third	Transpleural	Excision	Leiomyoma	Recovery
Kenworthy[23] Welch	1948	56	F	Anorexia; abdominal pain	Lower third Extended into cardia	Transpleural	Resection esophagogastrostomy	Leiomyoma	Recovery
Harrington[20]	1949	53	M	Cough; chest pain	Lower third	Transpleural	Excision	Leiomyoma	Recovery
Harrington[20]	1949	19	F	Choking; dysphagia	Lower third Extended into cardia	Transpleural	Resection esophagogastrostomy	Leiomyoma	Recovery
Chi[8] Adams	1950	25	M	None	Upper third	Transpleural	Resection esophagogastrostomy	Leiomyoma	Recovery
Goldman[18] Masters	1950	51	F	Dysphagia; substernal pain	Lower third	Transpleural	Resection esophagogastrostomy	Leiomyoma	Recovery

DISCUSSION

A survey of the literature discloses 39 benign intramural esophageal cysts and tumors surgically removed in 38 patients. The three cases herein reported bring the total of such lesions surgically treated to forty-two. A statistical analysis of so small a series is not justifiable, but several interesting aspects of these cases might be emphasized. The tumors were solitary in all but one instance in which two leiomyomas were found.[7] Twenty of the 42 tumors were located in the lower third of the esophagus, 15 in the middle, and 7 in the upper third. Twenty-six leiomyomas, eleven cysts, three fibromas, and two neurofibromas were encountered in this group of patients. There were 22 males and 19 females, with ages ranging from 19 months to 66 years. Three deaths were recorded in this series, and the result not given in one patient.

PATHOLOGY

The majority of benign intramural esophageal tumors are leiomyomas. These tumors arise from smooth muscle of the esophageal wall, are occasionally multiple, and are most commonly located in the lower third of this organ. They tend to remain localized to an arc of the esophageal wall, but in growth may partially or completely encircle it. Histologically the characteristic muscle bundles with a variable amount of fibrous stroma are seen. Metaplasia with striated muscle is occasionally noted.

Cysts of esophageal and bronchial origin are the second most common benign intramural esophageal tumors, being present in eleven of this series of cases. Embryologically they have a close relationship, and histologically their lining mucosa shares common characteristics which make their differentiation difficult. Since the embryologic esophagus has a ciliated epithelium, cysts of either origin may have a respiratory-like mucosal lining. The single distinguishing characteristic microscopically is that whereas bronchogenic cysts may contain cartilage, true esophageal cysts contain none of this tissue.

Olken[30] states that true esophageal cysts form a rather distinct group since they are almost invariably found in adults and are situated in the lower one-half of the esophagus. Bradford[4] elects to differentiate true esophageal cysts from bronchogenic cysts by their location, designating all those occurring in the esophageal wall as of esophageal origin and those elsewhere in the mediastinum as of bronchogenic origin. It seems reasonable to conclude that differentiation of these two varieties of cysts taking origin from so closely allied embryologic structures is extremely difficult if not impossible.

Three fibromas and two neurofibromas also occurred in this series of cases.

DIAGNOSIS

The literature suggests that the majority of benign esophageal tumors, especially those of fibromuscular origin, are asymptomatic. Statistics designed to indicate the incidence of benign esophageal tumors show the number of benign tumors found in patients complaining of dysphagia to be less

than that found in patients who do not have this complaint.[27] In this group of patients only four (all leiomyomas) had no symptoms, and their tumors were demonstrated as an incidental finding. Ten of twenty-six patients with leiomyomas had dysphagia as their chief complaint. One-half of the total number of patients had a chief complaint of dysphagia. Fifteen patients complained of pain in the chest described as "substernal distress," "tightness," or "fullness." Nine patients had respiratory complaints characterized by cough, dyspnea, choking sensations, and wheezing. Fourteen patients volunteered symptoms of epigastric burning, vomiting, and indigestion.

Significant physical findings were absent in this group of patients. The diagnosis of these tumors rests on contrast roentgenographic and fluoroscopic examination of the esophagus. The characteristic roentgenographic findings of benign intramural esophageal tumors have been well described.[19, 33] The demonstration of a smooth dome-shaped cap of barium adjacent to a rounded protrusion into the lumen of the esophagus suggests a benign intramural tumor.

Esophagoscopy should be done in each case and serves chiefly to allow a gross evaluation of the esophageal mucosa overlying the tumor. Biopsy by this route is usually of no value since, with the exception of tumors located near the cardia which may ulcerate, the overlying mucosa is usually intact. Indeed, biopsy through the intact mucosa is dangerous and may jeopardize easy enucleation by introducing infection into the tumor bed.

TREATMENT

With the exception of the relatively uncommon intramural lesions of the cervical esophagus, the transpleural route to these tumors is preferred. An approach from the left side is necessary in those lesions requiring resection of the esophagus. The right side may be utilized for tumors in which enucleation or limited excision of the mucosal bed will suffice. Thirty-eight of these 41 cases were approached transpleurally and one each by the abdominothoracic, abdominal, and cervical routes.

In the majority of instances, simple enucleation of the tumor may be accomplished without entering the lumen of the esophagus. This procedure was carried out successfully in thirty instances in this series (two tumors in one patient). In one patient so treated the result was not recorded, and one patient, a poor operative risk, died following an attempted enucleation of a fibroma of the upper third of the esophagus. Enucleation was uniformly successful in the cystic lesions. Following enucleation, the mural esophageal defect may be repaired by closure of the muscle or covered with a pedicled lung graft if extensive.[28]

Less frequently, the tumor may be attached so firmly to the mucous membrane that enucleation is impossible. In such situations, small portions of the tumor-bearing mucosa may be necessarily excised with closure of the defect. More extensive lesions require resection of the esophagus with esophagogastrostomy. In ten instances in this series some form of excision or resection

TABLE IV. MISCELLANEOUS ESOPHAGEAL TUMORS REMOVED BY ENUCLEATION

AUTHOR	DATE	AGE	SEX	SYMPTOMS	LOCATION	APPROACH	TREATMENT	DIAGNOSIS	RESULT
Churchill[9]	1937	19	F	Substernal pain; dysphagia	Middle third	Transpleural	Enucleation	Neurofibroma	Recovery
Barrett[2]	1940	66	F	Cough; dyspnea	Upper third	Transpleural	Attempted enucleation	Fibroma	Died
Adams[1, 25] Hoover Lahey	1945	48	F	Substernal distress; cough; dyspnea	Upper third	Cervical	Enucleation	Fibroma	Recovery
Cottet[13]	1949	32	F	Epigastric pain; dysphagia	Upper third	Transpleural	Enucleation	Fibroma	Recovery

was necessary. Three patients were treated by excision of a portion of the mucosa attached to the tumor, and seven patients required esophageal resection because of the tumor's extent. One of the three patients treated by excision died, and one of the seven patients requiring resection and esophagogastrostomy died.

It is conceivable that in small tumors requiring resection that end-to-end anastomosis of the esophagus might be accomplished, but no cases in this group were so treated.

SUMMARY

1. Three benign intramural esophageal tumors successfully treated by enucleation are reported.

2. The literature relative to benign intramural esophageal tumors is briefly reviewed.

ADDENDUM

Since this report was submitted for publication, another benign intramural leiomyoma located in the middle third of the esophagus has been successfully enucleated in this Clinic. The patient, a 15-year-old male (N.C.B.H. 125554), had a two-year history of intermittent dysphagia.

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THE PATHOLOGY OF EXPERIMENTALLY PRODUCED LYE BURNS AND STRICTURES OF THE ESOPHAGUS*

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THE modern treatment of lye burns of the esophagus and of the resulting strictures is founded on dilatation by a variety of methods. Early dilatation beginning several days after the fresh burn is enthusiastically supported by many authors,[1-6] and represents the initial phase of the "Salzer" method,[7] which is the accepted treatment in most clinics. The Jacksons,[8] however, recommend a delay of two to three weeks before starting dilatation.

In order to examine critically the rationale of this method of therapy, it seemed necessary to study the sequential pathologic changes resulting from lye burns. Because of the lack of any appropriate human pathologic material, we undertook to produce lye burns and strictures of the esophagus in the dog. Search of the literature revealed no previous successful attempt at the production of lye burns and strictures in the experimental animal. Certain observations made by Markow[9] from human material coincide roughly with our findings below.

METHOD

The initial experiments resulted only in failure. When lye solutions were introduced into the esophagus of the anesthetized animal in the erect position, the lye passed rapidly into the stomach and caused a fatal hemorrhagic gastritis within twenty-four hours. The outcome could not be prevented by aspiration and irrigation of the stomach, nor by the preliminary instillation of small amounts of hydrochloric acid. Balloon tamponade of the cardia proved unreliable.

In subsequent experiments laparotomy was performed under barbiturate anesthesia and the cardia temporarily occluded. In order to prevent aspiration of lye and to maintain adequate respiration the trachea was intubated at the start of the experiment and the endotracheal tube subsequently connected to a respirator. A large catheter connected to an available source of suction was inserted into the esophagus just below the cricopharyngeus level to prevent regurgitation of lye and to remove lye and irrigating solutions from the esophagus. With the animal suspended in a vertical position lye solution was introduced into the lower esophagus through a catheter.

Trial and error established the proper concentration of lye and duration of contact to achieve a sublethal effect: 10 to 15 cubic centimeters of a 10 per cent lye solution in contact with the esophagus for approximately sixty seconds. At the end of this period the lye was neutralized with a 0.1 normal hydrochloric

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acid and washed out with water. When concentrations of lye somewhat stronger than 10 per cent were used, perforations occurred frequently, up to the fifth day after the burn.

Animals maintained longer than five days after the burn were fed through a tubovalvular gastrostomy. No food or water was permitted orally.

EXPERIMENTAL RESULTS

The pathologic observations are based upon a study of fourteen animals in which the "standard" lye burn was produced (Table I). Nine animals were sacrificed or died from one to twelve days following the burn; the remaining five animals were maintained from four to eighteen weeks. All of these latter animals developed esophageal strictures (Figs. 3 and 4).

Fig. 1.

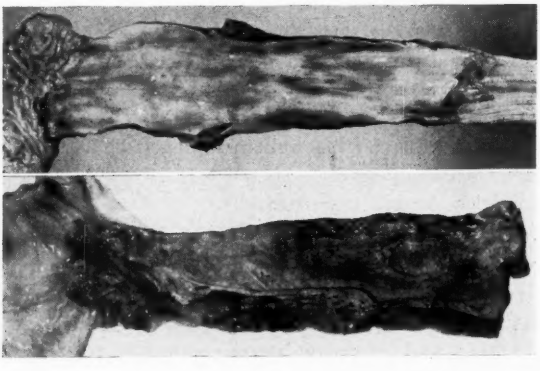

Fig. 2.

Fig. 1.—Forty-eight hours post burn. Specimen shows thickened, edematous wall with hemorrhagic areas. The superficial layers would undoubtedly have sloughed. On right note normal esophagus which was protected by aspiration catheter.

Fig. 2.—Four days post burn. Sloughing, superficial layers lifted freely away from underlying muscularis. Several ulcerations are apparent.

TABLE I

NO.	DURATION OF EXPERIMENT	REMARKS
27	24 hours	Sacrificed
28	48 hours	Sacrificed
12	48 hours	Sacrificed
23	4 days	Died from perforation and pneumothorax
16	5 days	Died from laryngeal edema and asphyxia
18	5 days	Died from perforation
11	7 days	Sacrificed
13	7 days	Sacrificed
22	12 days	Died; ? pylorospasm
14	4 weeks	Sacrificed
24	4 weeks	Died; ? pylorospasm
25	6 weeks	Sacrificed
21	13 weeks	Sacrificed
19	18 weeks	Sacrificed

Most animals manifested difficulty in swallowing saliva around the fifth to seventh day after the burn. This dysphagia progressed to a severe degree but was relieved between the fourth and sixth weeks in the three animals surviving that long. Since all strictures eventually proved to be incomplete, the temporary dysphagia for liquids was probably related in part to abnormal or decreased motility of the esophagus. This impression has not been checked with fluoroscopic studies.

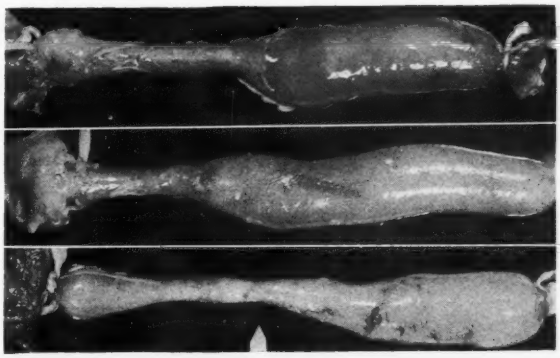

Fig. 3.—Lye strictures. Specimens distended with saline. Stomach to left. *Above:* One month post burn. *Middle:* Three months post burn. *Below:* Four months post burn.

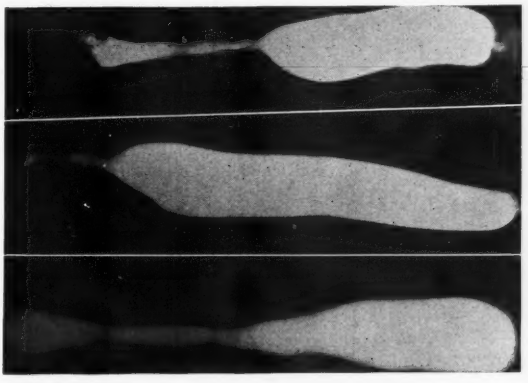

Fig. 4.—Lye strictures. Specimens same as in Fig. 3. Roentgenograms after injection with barium sulphate.

In the region of maximum burn, i.e., in the lower half of the esophagus, a constant series of pathologic events developed. At the end of twenty-four hours epithelial destruction was observed. Edema and congestion were apparent throughout the entire wall, but predominantly in the subglandular region of the submucosa (Fig. 6). By the end of forty-eight hours the submucosa was heavily infiltrated with inflammatory cells (Fig. 7). Many submucosal arteries and veins underwent thrombosis, and in this manner gangrene of the superficial layers became certain (Fig. 8).

Fig. 5. Fig. 6. Fig. 7.

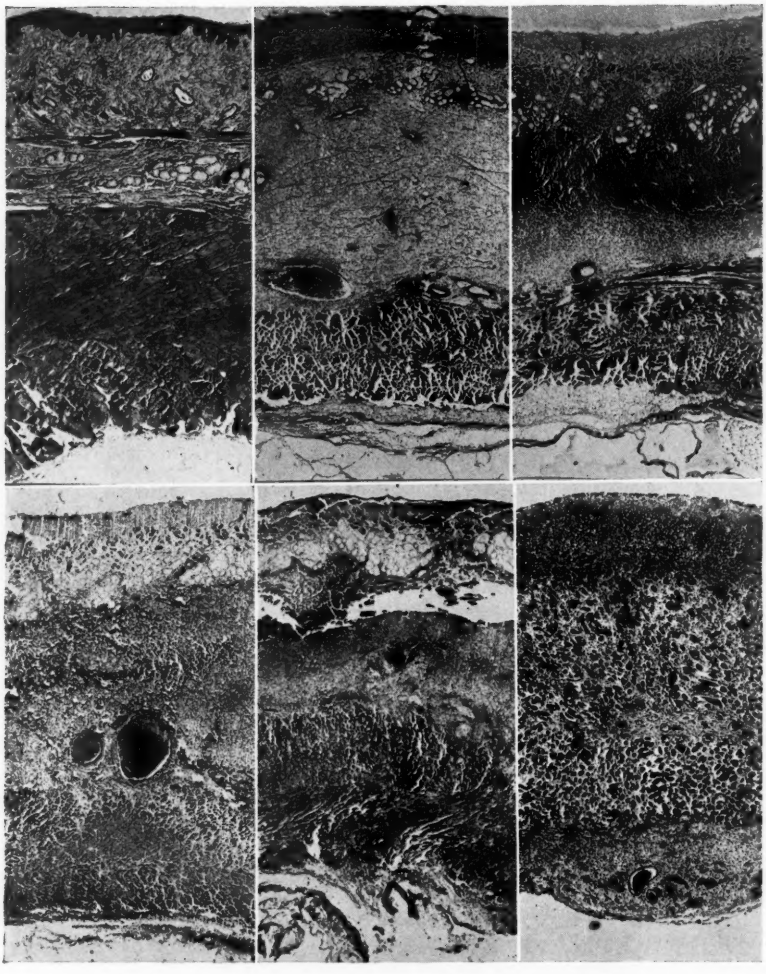

Fig. 8. Fig. 9. Fig. 10.

Fig. 5.—Normal esophagus. Note prominent muscularis mucosae, the location of the esophageal glands, and the relative thickness of the various zones.

Fig. 6.—Twenty-four hours post burn. Note absence of epithelium, generalized edema and congestion, especially marked in the submucosa, absence of cellular detail in the esophageal glands and in mucosal layer.

Fig. 7.—Twenty-four hours post burn. More advanced stage than in Fig. 6. Note heavy deposition of inflammatory cells in mid-portion of submucosa.

Fig. 8.—Forty-eight hours post burn. Thrombosed artery and vein deep in submucosa. Thrombosis is commonly seen at this stage.

Fig. 9.—Forty-eight hours post burn. More advanced stage than in Fig. 8. Liquefaction in region of intense inflammation with beginning separation of superficial layers.

Fig. 10.—Five days post burn. Mucosa and submucosa have sloughed leaving zone of inflammation and fibrin deposition internal to muscularis. Note considerable edema and some necrosis of muscularis with inflammatory zone external to muscularis.

A marked variation in the extent of musele necrosis and degeneration was observed in different animals. The impression was gained that the ultimate fate of these cells could not always be accurately predicted at this early stage. Certainly the magnitude of the edema, the advent of thrombosis, and the presence or absence of bacterial invasion were factors as important as the initial chemical insult in determining the fate of the muscularis.

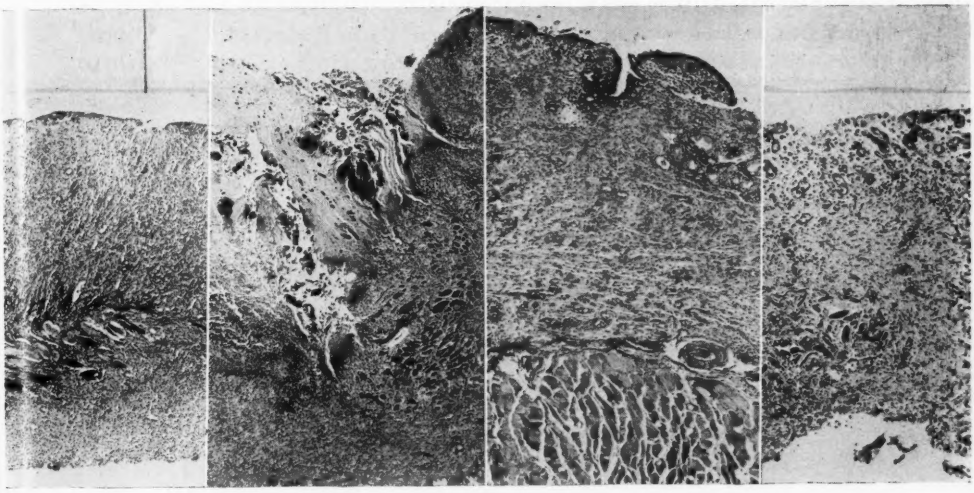

Fig. 11. Fig. 12. Fig. 13. Fig. 14.

Fig. 11.—Seven days post burn. Organization and fibrosis are proceeding rapidly. Section was taken through a region where muscularis was almost completely destroyed.

Fig. 12.—Seven days post burn. An intramural abscess showing liquefaction and bacterial clumps. Such evidence of frank bacterial inflammation has been observed not infrequently.

Fig. 13.—Seven days post burn. A less severely damaged region. The presence of esophageal glands and ducts and large submucosal vessels indicates that slough has not occurred. Note widening of submucosal zone, where fibrosis is in progress. Re-epithelization is taking place from duct.

Fig. 14.—Twelve days post burn. Organization somewhat delayed in this region of severe destruction. Note almost complete necrosis of muscularis.

As the inflammatory zone in the submucosa became more intense, liquefaction ensued at this level and the nonviable mucosa and submucosa sloughed (Fig. 9). This process was complete by the fifth day, and sometimes occurred sooner. Thereafter the residual inflammatory zone underwent rapid organization and a layer of fresh granulation tissue was soon formed internal to the muscularis (Fig. 10). Subsequently young fibroblasts flattened out along the internal surface and formed a false lining membrane. Fibrous tissue was deposited in the muscularis in proportion to the extent of damage there.

Variations in the above described pathologic process were sometimes observed. Rarely, destruction of the muscularis occurred very rapidly without the preliminary phase of marked submucosal edema and inflammation. This suggested a more serious chemical insult. After forty-eight hours it was not too uncommon to find evidence of bacterial invasion of the muscularis varying from a phlegmon to a localized intramural abscess (Fig. 12).

It is surprising that of the fourteen animals prepared by the "standard" burn method only two died from perforation. Mediastinitis was not observed since the perforations occurred directly into the pleural space with early death due to pneumothorax and purulent effusion. A protective layer of fibrin was usually deposited on the external surface of the esophagus as necrosis or inflammation involved the muscularis. In the absence of abscess formation organization and fibrosis followed immediately in the wake of the destructive processes and served to maintain the integrity of the wall (Fig. 11).

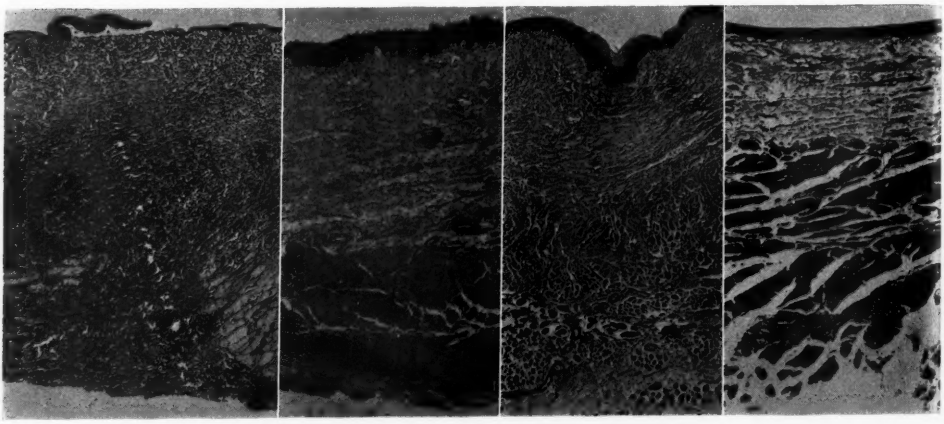

Fig. 15. Fig. 16. Fig. 17. Fig. 18.

Fig. 15.—One month post burn. Fibrosis extends through entire wall at this point. Note advancing epithelium lifted away from granulation tissue which it is attempting to cover.

Fig. 16.—Thirteen weeks post burn. From region of stricture. Note good preservation of muscularis, wide zone of fibrous tissue, absence of esophageal glands, and good epithelial layer.

Fig. 17.—Thirteen weeks post burn. From region of stricture. In this instance there is considerable fibrosis within the muscularis.

Fig. 18.—Eighteen weeks post burn. From region of stricture. Attenuation of wall probably related to method of fixation. Note changes in submucosa despite good preservation of muscularis. Observe thin strands of striated muscle fibers dispersed in fibrous zone.

In areas of less pronounced damage, i.e., in the upper half of the esophagus, frank sloughing of the mucosa and submucosa did not necessarily occur. Usually the epithelium was destroyed and fibrin deposited in the edematous mucosa and submucosa. Some of the glands lost their cellular detail. Residual ducts provided a source for early re-epithelization (Fig. 13).

When sloughing of the internal layers occurred, re-epithelization was not complete before the sixth week in our animals. The regenerated epithelium, at first very thin and flat, gradually thickened and eventually assumed an essentially normal appearance. Submucosal esophageal glands remained permanently absent, however. The granulation tissue matured only after epithelization had taken place, and only then did evidence of chronic inflammation disappear.

In one animal which succumbed four weeks after the burn, presumably from the effects of pylorospasm and vomiting, there was histologic evidence of

repeated irritation or trauma. Superficial necrosis and acute inflammation was noted in the layer of granulation tissue lining the esophagus. The important role of gastric juices is assumed, although not proved.

Stricture formation developed after the second week and before the end of the fourth week. From a study of the three animals sacrificed at six, thirteen, and eighteen weeks, respectively, it is apparent that marked strictures may develop in the presence of a relatively normal muscularis (Figs. 16, 17, 18). Evidence is not available from these investigations as to whether such experimental strictures are progressive. In this series of animals the effect of mechanical dilatation has not been tested.

COMMENT AND SUMMARY

1. A method has been described for the production of sublethal lye burns and strictures of the esophagus in the dog.

2. The pathologic events following a fresh lye burn in the dog are rather constant. They consist chiefly of edema and congestion, principally of the submucosal layer; inflammation of the submucosa and thrombosis of its vessels; sloughing of the superficial layers; necrosis of the muscularis in varying degrees; organization and fibrosis on the internal surface of and within the muscularis; and delayed re-epithelization.

3. Bacterial invasion may play a significant, although by no means a decisive role in the pathogenesis of the lye burn and stricture. For this reason antibiotic therapy should be given early and intensively.

4. Re-epithelization is a slow process, particularly where the slough has been extensive. Re-epithelization permits subsidence of chronic inflammation and maturation of the granulation tissue. Complete rest of the esophagus may be advisable to facilitate and hasten this phase of the repair, thus minimizing the production of scar tissue.

5. In view of the necrotic and inflammatory phase observed during the first week after the burn, early dilatation (i.e., before the tenth day) seems illogical and, if vigorously employed, almost certain to intensify the damage and delay reparative processes. Evaluation of delayed dilatation must await further investigation.

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CERVICAL ESOPHAGOSTOMY

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DURING the course of treatment of certain cancers of pharyngeal, facio-maxillary, and cervical areas, gavage feedings are required for extended periods of time. In certain cases, it is impossible to pass a feeding tube through a site of obstruction in the pharynx or upper esophagus. In others, the tube must remain and its presence becomes extremely irritating; a situation which may be further aggravated if periodic changing of the tube is not well tolerated. Under such conditions, there has often been constructed for feeding purposes a gastrostomy which, even under the best of conditions, is not too satisfactory.

As a pharyngostomy had been found to constitute a satisfactory port of entry for a feeding tube when such an opening had been created in the course of removal of the larynx and portions of the pharynx, a cervical esophagostomy might be a solution to the feeding problem in those cases in which the site of obstruction was above the lower end of the cervical esophagus. Such a procedure has been employed successfully in one patient. As a survey of recent reference books and current literature fails to disclose a description of the use of this operative procedure for this specific purpose, this case report seemed indicated.

CASE 1.—This 47-year-old woman was admitted to Episcopal Hospital in September, 1950, with an established diagnosis of epidermoid carcinoma of the cervical esophagus, and right lower pharyngeal wall. She had refused surgical removal of the lesion, and had received three x-ray treatments to her neck in the previous three days, and the partial obstruction of her cervical esophagus had become complete. It was decided to perform a cervical esophagostomy rather than a gastrostomy, for feeding purposes.

Operation: On Sept. 16, 1950 (under local anesthesia), a transverse incision was made just above the clavicle over the lower border of the sternocleidomastoid muscle. In the face of the poor prognosis, and in order to simplify exposure, the insertion of the sternocleidomastoid muscle was divided exposing the internal jugular vein. The jugular vein, common carotid artery, and vagus nerve were retracted laterally exposing the lateral wall of the cervical esophagus. The inferior thyroid artery exposed in the depth of the wound was ligated. Traction sutures were placed in the esophageal wall, and a small opening made into the lumen. A Levine tube was passed through this opening down the esophagus into the stomach (Fig. 1). This was anchored by a purse-string suture at the site of entrance into the esophagus and by a suture at the skin margin. A petroleum jelly gauze pack was placed between the tube and great vessels of the neck. The portion of the incision overlying the great vessels was sutured, but the remainder remained open.

Postoperatively, she was given 300,000 units of penicillin (crysticillin) daily for six days. Nothing was given through the tube for twenty-four hours, after which time a 3,000 calorie liquid diet was begun. Drainage from the neck wound was minimal, and consisted of a serous exudate. Temperature remained normal and no induration developed about the incision.

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By the time of discharge in six days, she gave her own feedings with no difficulty. By this time she was able to swallow saliva, but this did not increase the amount of drainage from the wound. Two weeks later, healing had progressed until only a well-defined, small sinus tract remained. The Levine tube could easily be removed and a new one reinserted, but it was considered best to keep the tube in place continuously, except when being changed, in order to prevent closure of the sinus tract.

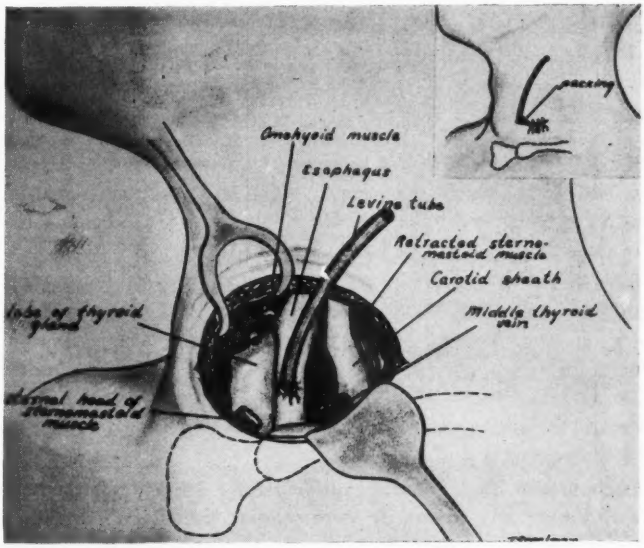

Fig. 1.—Cervical esophagostomy.

DISCUSSION

Technically, this procedure is a simple one. It can be done under local anesthesia in a few minutes, with little discomfort to the patient. Any contamination of the wound is readily handled by use of antibiotics and omission of closure of the incision minimizes the possibility of spread of any infection into the superior mediastinum.

Care of the esophagostomy is simple. Drainage is minimal, and regurgitation or leakage of prepared or partially digested food does not occur. Skin excoriation is not present. The tube can be concealed by a simple dressing, can be easily changed, and is accessible for feeding purposes without the necessity for removal of clothing or use of a recumbent position.

In a patient who requires gavage feedings for an extended period of time and who cannot pass his own feeding tube easily, or who has complete obstruction of the food-way at a sufficiently high level that cervical esophagostomy can be carried out below the site of obstruction, its use would seem to be the procedure of choice.

THE MOBILIZATION OF THE ARCH OF THE AORTA IN ORDER TO REACH THE BIFURCATION OF THE TRACHEA

OLIVIER MONOD, M.D., G. BABOU, M.D., E. KATEB, M.D., AND S. GHAZI, M.D., PARIS, FRANCE

FROM the left side, it is difficult to reach the origin of the left main bronchus and the end of the trachea, especially when the lung has been removed and the mediastinal tissues are inflamed. In such cases we propose to mobilize the arch of the aorta and to retract it temporarily, inferiorly and to the left.

We were forced to have recourse to this procedure in the case of a wide fistula which resulted from the removal of the left lung because of tuberculosis. This gave us an astonishing freedom of action. The left bronchus having been resected near its origin, the bronchial fistula had retracted far back under the arch of the aorta. The dissection of the fistula in the concavity of the aorta had been made impossible by inflammatory sclerosis and the resistance of the tissues and the organs.

We encountered two technical difficulties during the operation. First, there was no longer any pleura or loose cellular tissue around the aorta. Dissection was laborious and nerve-racking. Second, the particular fragility of the intercostal arteries at their origin made their isolation and section especially delicate.

Mr. D. (born 1928) was first examined in 1947. In 1948, a left intrapleural pneumothorax and an intrapleural pneumolysis were performed. The following year, purulent pleurisy with perforation necessitated a pleurotomy.

In January, 1950, there developed a severe pyothorax with a high temperature and a high sedimentation rate. The pulse rate was 96 and blood pressure 110/70. Bronchoscopy (Aug. 9, 1949) showed normal bronchi. Roentgenograms of the right lung had always been normal. The patient was given 116 Gm. of streptomycin during 1948.

A thoraco-pleuro-pneumonectomy was performed on Jan. 31, 1950. The chest wall (seven upper ribs and spaces) was removed at the same time as the whole of the parietal pleura. The three pulmonary vessels were divided and sewn. The bronchus was closed by a clamp and its walls sewn together by mattress stitches. The crushed part was resected, and a terminal suture made by means of separate stitches (silk 000). The whole was then covered with a flap of the pericardium, which covered it well. Closure was made without drainage.

The postoperative course was normal, with the temperature returning to normal after the third day. The patient regained his preoperative weight after two weeks. There was no sputum. The patient left the sanatorium on May 2, 1950.

At the end of May, four months after the operation, the patient became feverish and suffered from dyspnea, and the bronchial fistula made itself evident. The parietal scar reopened.

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Bronchoscopy.—Bronchoscopy was performed on July 8, 1950. In the left bronchus, at 1 cm. from the carina, a fistula was seen on the left edge of the bronchial scar.

In October, 1950, the patient began to cough, and to expectorate, and air could be heard passing through the fistula on both inspiration and expiration. There was a residual thoracic cavity with a wide opening to it in the back.

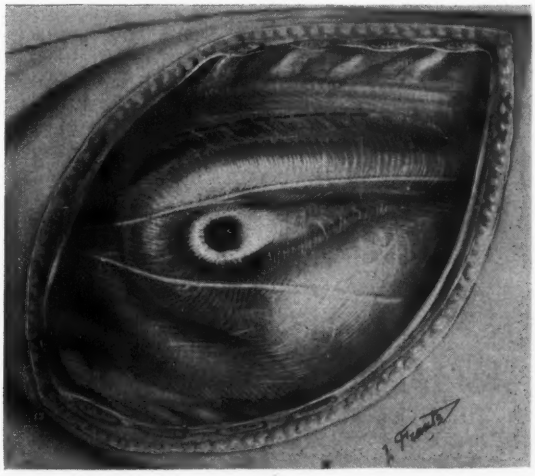

Fig. 1.—Left thoraco-pleuro-pneumonectomy. Fully opened fistulization of the left bronchial stump divided at its origin. The thorax, opened, shows the arch of the aorta. The uninterrupted line shows the incision for the freeing of the aorta.

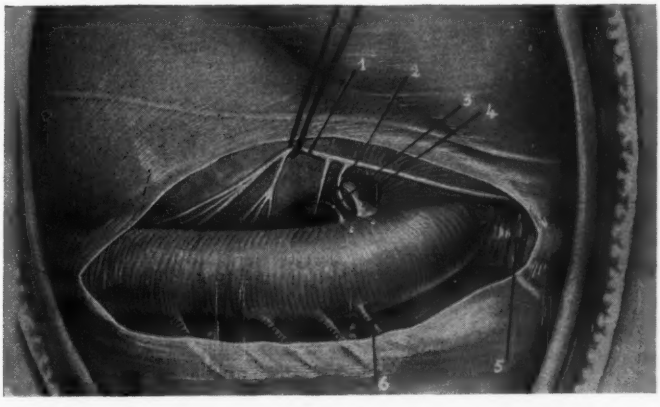

Fig. 2.—The dissection of the aorta. *1*, The vague nerve. *2*, The recurrent nerve. *3*, The right bronchial artery. *4*, The arterial ligament. *5*, The left superior intercostal vein. (It is generally situated lower, along the aorta.) *6*, The fourth left intercostal artery.

Myoplasty.—Oct. 18, 1950. The fistula was situated just in the concavity of the aorta arch and was deeply seated. It was plugged with muscle with a piece of placenta over it. The cleaning up of the fistula had been done in a summary and unsatisfactory fashion. It had been impossible to reach the wall of the bronchus for lack of space through which to get at the concavity of the aorta.

The symptoms of fistula reappeared immediately. It was decided to free the aorta so as to reach the fistula.

Mobilization of the Aorta for the Suturing of a Bronchial Fistula.—On Nov. 7, 1950, by means of laborious and difficult dissection, through tissues that were inflamed, infected, and scarred, the arch of the aorta and the beginning of the descending aorta were eventually freed, from the left subclavian artery as far as the seventh intercostal space. The intercostal arteries in the fourth, fifth, sixth, and seventh spaces, arising from the posterior surface of the origin of the thoracic aorta were divided. As their walls were very thin and fragile, they could not be stretched, and great care was required in freeing and dividing them. A right bronchial artery which arose near the arterial ligament was also divided. The ligature and dividing of the arterial ligament gave an appreciable increase of mobility. The recurrent nerve was located and avoided. Once the aorta had been freed, rubber bands were put round it to displace it downward and to the right. The upper part of the vessel dilated under the action of the blood, but care was taken not to interrupt the circulation completely. The trachea was reached and the edges of the bronchial fistula were dissected out. It was noted that the stump of the left bronchus was fully open.

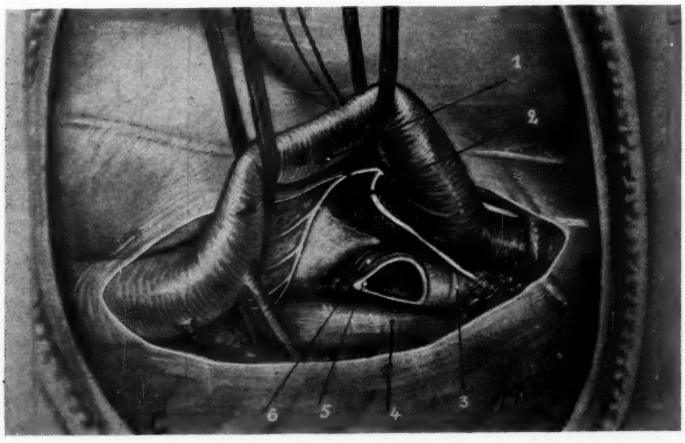

Fig. 3.—The "bucket handle" mobilization of the aorta. *1*, Section of the intercostal arteries. *2*, The recurrent nerve. *3*, The left side of the terminal trachea. *4*, The esophagus. *5*, The carina. *6*, The stub of the right bronchus.

The dissection of the fistula and the cleaning up of the edges of the bronchus were laborious. The suture was made in healthy tissue under very satisfactory conditions by separate stitches with silk No. 00. There now existed practically no bronchus, for it had been cut almost on a level with the trachea during the first operation and during the cleaning up. It was, therefore, possible only to make a terminal suture by a single row of stitches.

The work of dissection, cleaning up, and suture had been done partly in the concavity of the aorta, this vessel being turned right back, the vagus nerve lying anteriorly, and partly by turning the aorta toward the right, the operator working from behind the patient. The suture was covered with cellular tissue in a satisfactory manner. The aorta, released, returned automatically to its place and pressed against the suture.

Postoperative Course.—There has been no cough or expectoration since the operation, the dyspnea has diminished, and the air no longer enters the left residual thoracic cavity.

TRACHEOTOMY, A USEFUL PROCEDURE IN THORACIC SURGERY, WITH PARTICULAR REFERENCE TO ITS EMPLOYMENT IN CRUSHING INJURIES OF THE THORAX

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ALTHOUGH tracheotomy is one of the oldest known surgical procedures, its usefulness in thoracic surgery has not been hitherto emphasized, nor have its physiologic effects upon the respiratory mechanism been adequately investigated. Our interest in tracheotomy was stimulated by the strikingly successful results of its use two years ago in the treatment of two patients with severe crushing injuries of the thorax. This procedure has continued to produce such satisfactory improvement in patients with this type of injury that an investigation was undertaken to discover how it influences the physiologic status of patients in respiratory distress from various causes.

The effects of tracheotomy fall into two main categories; mechanical and physiological. The former comprises: (1) the ability to maintain a clear airway by easy removal of excessive fluids or blood from the tracheobronchial tree, and (2) the relief of laryngeal obstruction resulting from edema, bleeding, or faulty functioning of the vocal cords. Scrutiny of the literature has failed to reveal mention of the effects of tracheotomy on the physiology of respiration. Such effects are of real importance and we believe the principal ones to be: (1) a decrease in the amount of dead space in the respiratory tree with a resulting increase in effective ventilation, and (2) a decrease in resistance to breathing, both on inspiration and expiration.

Before discussing our clinical and laboratory investigation, it appears desirable to state the problems presented by crushing injuries of the thorax and to present case reports illustrating the results obtained with tracheotomy. "Crushed chest," "flail chest," and "stove-in-chest" are indefinite but descriptive terms applied to a variety of nonpenetrating thoracic injuries characterized by multiple rib fractures which cause movement of a portion of the thoracic wall paradoxical to the normal excursions of the thoracic cage with mediastinal flutter. These injuries frequently result from severe compression, and are often associated with trauma to the extremities, head, or viscera of the thorax or abdomen, in variable combinations. Consequently, categorical analysis of this type of injury is difficult. A multiplicity of complications including hemothorax, simple or tension pneumothorax, and "wet lung" may predominate in a particular case and demand accurate supplementary therapy. There is,

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however, a uniformity in the clinical picture of this type of injury. Respirations are short and rapid and are accompanied, in the conscious patient, by great pain and marked apprehension. Cyanosis, high respiratory rate, and tachycardia further indicate respiratory distress. Bony crepitus and paradoxical movement of the thoracic wall in the affected area indicate segmental fractures of ribs. The tracheobronchial tree, initially dry unless the patient has associated bronchial or pulmonary trauma, may become filled with secretions several hours after the injury. Brewer and his co-workers[3] recognized this condition as a clinical entity and termed it "wet lung." A vicious circle is established by these two factors. The cyanosis and breathing effort resulting from paradoxical respiration cause the wet lung syndrome which, calling for greater effort, results in more marked paradoxical movements of the loose segment of the thoracic wall. Because of pain, cough is ineffectual, and plugging of the alveoli and bronchi further impairs mechanical ventilation of the lung.

This injury is attended by a high mortality. That most forms of therapy are inadequate is evidenced by the number and variety of recommended measures. A brief review of those most commonly proposed illustrates the confusion prevailing in treatment of this condition. Dolley and Brewer[8] recommended (1) morphine in doses large enough to control pain, (2) strapping of the thorax with adhesive, and (3) aspiration of the trachea by catheter. Blades,[2] on the other hand, emphasized the dangerous reduction in vital capacity following strapping of the chest. Intercostal nerve block as a means of controlling pain was first advocated by Italian surgeons in World War I[7] and won popular acclaim during World War II.[5, 9, 10, 18] Numerous methods of fixation of the loose segments of the thoracic cage have been proposed, such as the use of screw hooks,[13, 14, 15] tenaculae,[4, 5, 12, 17] or wire,[10, 16, 18] with or without balanced traction, in the ribs or in the sternum, or in both. The disadvantages of these methods lies in the fact that mechanical fixation is difficult in constantly moving cancellous bone. Coleman[6] has recently recommended that each fractured rib be immobilized by individual wiring and pegging, a method which requires extensive surgery in critically injured patients. Administration of oxygen under positive pressure has been found beneficial, but it does not produce striking improvement. Hagen[11] has reported the use of the Drinker respirator.

The principal aims of therapy for "crushed chest" are to eliminate paradoxical motion as a factor in inadequate respiration, to keep the tracheobronchial tree free of secretions, and to relieve pain. Although intercostal nerve block has been successful in the control of pain, no commonly accepted method of eliminating paradoxical respiration has been reported. Tracheotomy, which has hitherto been emphasized in the literature for the relief of laryngeal obstruction, has recently been advocated as a useful adjunct in keeping the tracheobronchial tree free of secretions in patients with severe thoracic trauma.[1] In our experience, tracheotomy has been useful in facilitating maintenance of a clear airway, and this is one of its important advantages. In addition, the paradoxical excursions of the loose segment of the thoracic wall have been

markedly decreased in each patient with crushed chest treated by tracheotomy in this clinic. Pain has been strikingly relieved, probably as a consequence of lessened effort in respiration which in turn results from the diminished movement of the loose segment and from the decreased resistance to inspiration and expiration. As a result oxygenation has been markedly improved even though there was no evidence of laryngeal or tracheal obstruction or of excessive bronchial secretions.

CASE REPORTS

CASE 1.—L. K. (246807). This 58-year-old white man was admitted on Jan. 11, 1949, after being in an automobile accident. He complained of pain all over the anterior aspect of the thorax and over the sacrum posteriorly. Roentgenological examination revealed a fractured left pelvis involving the superior and inferior rami, and a fracture of the transverse process of the fifth lumbar vertebra on the left. Blood pressure was 120/90; pulse 116 at the radial artery and 130 at the apex; respirations 36. He was obese, with rapid short respirations, conscious, and cooperative. He was receiving oxygen per nasal catheter and his color was satisfactory. Examination of the thorax showed bony crepitus of the anterior wall bilaterally, involving the upper ribs. There was pain on pressure over the sternum. No paradoxical respirations were recognized at this time; the thorax was clear to auscultation. The heart sounds were irregular with an apparent pulse deficit. Roentgenograms showed fractures of ribs 3,4,8,9 in the anterior axillary line on the right and of 3,4,6 in similar position on the left. In the next twelve hours his respirations became increasingly difficult, dyspnea and pain more marked, and cyanosis appeared despite oxygen. Paradoxical movement of the anterior thoracic wall now became noticeable. His tracheobronchial tree, however, remained clear. A tracheotomy was done at this time and although aspiration failed to produce any significant amount of secretion, the procedure produced a great deal of respiratory relief. His respirations were much less labored and the sucking motion of his thoracic wall nearly ceased. His color was immensely improved and apprehension disappeared. His progress thereafter was steady. The tracheotomy tube was removed sixteen days later.

CASE 2.—H. K. (246896). This 61-year-old white man was admitted on Jan. 15, 1949, following an automobile accident. He was unconscious on admission. The left pupil was slightly larger than the right, and Babinski test was positive on both sides. All reflexes were active and equal bilaterally. Severe cyanosis was present; blood pressure was 125/90. He was given oxygen by mask, and within an hour began to regain consciousness, shortly becoming awake and responsive, but unable to talk except in gutteral tones. Respirations were jerky, predominantly diaphragmatic, and he was markedly cyanotic when not receiving oxygen. There was bony crepitation in the region of the upper four ribs in the costochondral area on the right. Respirations were paradoxical in this area. The chest was clear to auscultation, and breath sounds were equal bilaterally. There was an acromio-clavicular separation on the right and an irregular dirty laceration of the right buttock through which a large comminuted fracture of the sacrum protruded. The latter wound was hastily debrided under local anesthesia and the wound left open. His condition rapidly began to deteriorate. Respirations became labored and he had to be restrained because of excitability. Examination now revealed considerable subcutaneous emphysema of the right thorax and neck. Blood pressure was 120/90; respirations 36, pulse 108. Thoracentesis on the right produced 1,000 c.c. of air which was not under tension, but no particular improvement followed this procedure. The patient steadily grew worse until he was considered to be in extremis. Tracheotomy was performed with immediate and spectacular improvement. Respirations became quiet and easy, excursions diminished, his color changed from cyanotic to normal, restlessness disappeared, and he was without pain. Crepitation lessened, the emphysema ceased to extend, and his improvement was slow but steady. Oxygen was discontinued two days later, but digitoxin was necessary because of the patient's development of fibrillation. The tracheotomy tube was removed on the fourteenth hospital day, and he was discharged on Feb. 14, 1949.

Comment.—In both of these elderly patients, tracheotomy was followed by a prompt decrease in paradoxical motion of the loose segment of the thoracic wall, marked improvement in color, and rapid relief of respiratory distress and apprehension. Since in each case the respiratory tree was clear before tracheotomy, improvement could not be explained on the basis of removal of secretion or relief of obstruction in the usual sense.

CASE 3.—L. M. N. This 53-year-old white man was admitted on May 2, 1949. He was alcoholic on admission, and police reported that he had driven his car into a streetcar. He was a well nourished white man, only partially cooperative, complaining of pain in his chest. There was marked respiratory difficulty, but no paradoxical motion and no cyanosis. The trachea was in midline. There was crepitation in the anterior axillary line on both sides with a minimal amount of subcutaneous emphysema. It was difficult to determine how many ribs were fractured. His lungs were clear to auscultation. Respirations were diaphragmatic. Roentgenograms showed fracture of ribs 4 and 5 on the right and of 3,4,5,6 on the left. His respiratory difficulty increased in the following forty-eight hours, in spite of oxygen therapy. Paradoxical motion became apparent and his thorax became filled with coarse rhonchi. He became markedly cyanotic and dyspneic and the trachea shifted to the left. Tracheotomy was performed and a large amount of mucus was aspirated from the tracheobronchial tree. Respirations were noted to become easier immediately. Aspiration continued to be necessary at frequent intervals. He did well thereafter. The tracheotomy tube was removed on the eleventh hospital day, and the patient left the hospital against advice on the following day.

Comment.—Paradoxical respiration was not evident in this instance until the wet lung syndrome occurred with the concomitant increased respiratory effort. Undoubtedly, the accumulation of tracheobronchial secretions was the important factor which tracheotomy was particularly useful in relieving. The importance of tracheotomy in these circumstances cannot be overemphasized. It obviates the need for repeated bronchoscopy, and permits frequent and simple aspirations of the tracheobronchial tree to be performed by the nursing staff. Bronchial secretions which otherwise stagnate can be removed, and the complications of atelectasis and consequent pneumonia can be averted.

CASE 4.—A. K. (235588). This 50-year-old white woman was admitted to the fracture service on April 13, 1949, after having fallen from a second story porch. She complained of severe pain in the lumbar area and on both sides of her chest. Physical examination revealed an agitated white woman who was somewhat confused. Pulse was 90; respirations 60; blood pressure 125/80. She was treated supportively. Subsequent examination revealed multiple rib fractures: 2 through 8 on the right and 5 through 10 on the left side. She also had a compression fracture of the fourth lumbar vertebra. Paradoxical respirations became apparent and increased in violence during the next few days. She was given oxygen by nasal catheter, and became dyspneic whenever oxygen was discontinued. She was treated with intratracheal aspiration which was productive of moderate amounts of material. Repeated bronchoscopy produced amounts of thick material varying from moderate to large and was followed each time by temporary improvement. A tracheotomy was perfored on the tenth hospital day and a large amount of mucus evacuated. Her respirations remained labored, and she died twenty-five hours later. Necropsy revealed pulmonary congestion and edema, slight atelectasis of the left lower lobe, extensive hemorrhage into the adrenals, moderate hemoperitoneum, and focal laceration of the spleen, in addition to her thoracic injury. Pathologists concurred in the clinical impression that the patient died from aspiration pneumonia following crushed chest syndrome.

Comment.—According to our present standards, this patient was treated improperly. She obviously had a severe thoracic injury which should have been treated by prompt tracheotomy in order to provide improved ventilation by control of paradoxical movement and by thorough clearing of the tracheobronchial tree.

CASE 5.—L. S. (262207). This 45-year-old white woman was admitted to the receiving ward on Mar. 26, 1950, after having been involved in an automobile-train accident. She was comatose on admission, breathing rapidly with grunting respirations, but she moved all extremities, was moderately restless, and responded to painful stimuli. Her pulse was 128, blood pressure 90/60, and respirations 44. Her skin was pale and cool, and there was some cyanosis which became less noticeable after administration of oxygen. The trachea was in the midline. Thoracic examination revealed subcutaneous emphysema over the entire thorax anteriorly and extending into the neck. Palpation revealed bony crepitation along the right side of the sternum in the region of the second, third, fourth, and fifth ribs. Auscultation revealed no rhonchi. Aspiration of the right pleural cavity produced 10 c.c. blood and 50 c.c. air. Roentgenograms showed anterior and posterior fractures of ribs 2 through 5 on the right side. There was marked paradoxical movement of a large portion of the right thorax, particularly in the sternal area. Tracheotomy was performed, and immediate improvement was evident. The color of the skin became progressively better, breathing was rapid but easy, and the subcutaneous emphysema was lessened. Her course was generally good. There was notable absence of pain and apprehension. She periodically accumulated moderate amounts of mucus in the tracheobronchial tree which required aspiration. The tracheotomy tube was removed on the fourteenth day, and she was dismissed soon thereafter.

CASE 6.—W. L. (268913). This 47-year-old Negro man was admitted to the receiving ward on Oct. 29, 1950 after having been in an automobile accident. Examination revealed a crushing injury of the thorax and a severe head injury. The ribs were fractured in three places: right, 1,2,3,4, in the mid-clavicular line; left, 3,4,5,6,7 in the anterior axillary line; and left, 3 through 9 in the mid-scapular line posteriorly. He was unconscious, with a hemiplegia on the left side. Lumbar puncture showed a pressure of 126 mm. H_2O. Spinal fluid contained 10,000 red blood cells per cubic centimeter. Respirations were short, shallow, and paradoxical. He was cyanotic in spite of oxygen. His condition on admission was so poor that it was doubtful whether he would survive the night. Tracheotomy was performed, however, and his respirations immediately became easier and paradoxical movement diminished noticeably. His neurologic status, involving severe midbrain damage, deteriorated rapidly, and he expired four days after admission. Necropsy revealed marked acute cerebral edema with encephalomalacia. He also had pulmonary congestion and a small (500 c.c.) hemopneumothorax on the right.

Comment.—In each of these two cases there was an immediate improvement in oxygenation following tracheotomy. The paradoxical movement of the loose segment of the thoracic cage was noticeably lessened, pain was relieved, and the tracheobronchial tree kept clear of excessive secretions. The color improved almost immediately and the agitation and apprehension disappeared. In Case 5 studies were made of the oxygen saturation of the arterial blood before and after tracheotomy. Studies of intrapleural pressure were made in both cases. These will be discussed subsequently.

It is important to point out that in the majority of cases observed, the tracheobronchial tree was clear, so that the usually accepted purpose of tracheotomy, i.e., the relief of obstruction and the removal of secretions, was not the primary result of this procedure. Obviously, additional factors were involved.

The manner by which the marked respiratory improvement was effected suggested directions for laboratory and clinical investigation. During the past year, studies have been made to clarify some of the physiologic relationships in respiratory embarrassment. Our knowledge is not yet as complete as we should like, but we have corroborated our initial empirical observations.

Pulmonary function resolves itself into the ventilatory function, which is the mechanical movement of air in and out of the lung, and the respiratory function by which is meant the diffusion of gases across the alveolar membrane. The influence of tracheotomy primarily concerns the former. Consideration of the effects of this procedure upon the ventilation component of pulmonary function suggests that two are of importance. These are (1) the reduction in volume of the dead space involved in the respiratory act, i.e., that of the nasal passages, mouth, pharynx, trachea, and bronchial tree, and (2) the decrease in the resistance to the inspired and expired air.

By changing the point of entrance and exit of respired air from the nose and mouth to the trachea just above the suprasternal notch, the dead space is reduced about 100 c.c. (Fig. 1). This figure was determined by observations made on cadavers. The esophagus was ligated at its origin from the hypopharynx and the mouth-trachea-pharynx was filled with water to the point of the usual tracheotomy opening. *With the mouth opened in a position corresponding to easy mouth breathing,* this volume was found to be 110, 115, and 125 c.c. in three recently deceased men. Though the figures are not precise, they indicate that a substantial decrease in dead space is effected by tracheotomy. This decrease in dead space contributes to the more effective utilization of the tidal air whether it is of a normal or diminished amount. In instances of crushed chest, the tidal air is decreased as a result (1) of the interruption of the efficiency of the thoracic bellows by the paradoxical movement of a segment of the thoracic cage and (2) of the voluntary diminution in respiratory effort which results from the severe pain produced by the movement of the fractured ribs. Additional factors producing respiratory embarrassment are wet lung, pneumothorax, especially if under tension, hemothorax, and pulmonary injury.

Attempts were made to determine the tidal air pre- and post-tracheotomy of dogs in which segmented fractures of six ribs had been produced by operation. It was found that the anesthetized animals tolerated this procedure so well that it was impossible to produce the complete crushed chest syndrome in them. Further, determination of tidal air in such animals after tracheotomy by using the Collins spirometer was extremely unsatisfactory because of the failure to eliminate leaks or obstructions in the tracheal tube attachments and face masks. Determinations of tidal air were made, however, in human cases 5 and 6 and were found to be 320 c.c. and 330 c.c. in comparison with the commonly accepted figures of 500 c.c., whereas in two patients with less severe thoracic injuries the tidal air was found to be 500 c.c. and 540 c.c., respectively. The more effective utilization of a tidal air reduced to levels incompatible with adequate ventilation by diminishing the volume of the dead space appeals to us as an important physiological effect of tracheotomy.

It seemed logical to believe that tracheotomy, since it bypasses the nasal passages, pharynx, larynx, and a segment of the trachea, would decrease the resistance offered to the inspired and expired air, and that this should result in more efficient pulmonary function. Any such reduction in resistance should be reflected in the intrapleural pressure. Accordingly measurements of intrapleural pressures were determined both in experimental animals and in human patients before and after tracheotomy. Mongrel dogs weighing 15 to 20 kilo-

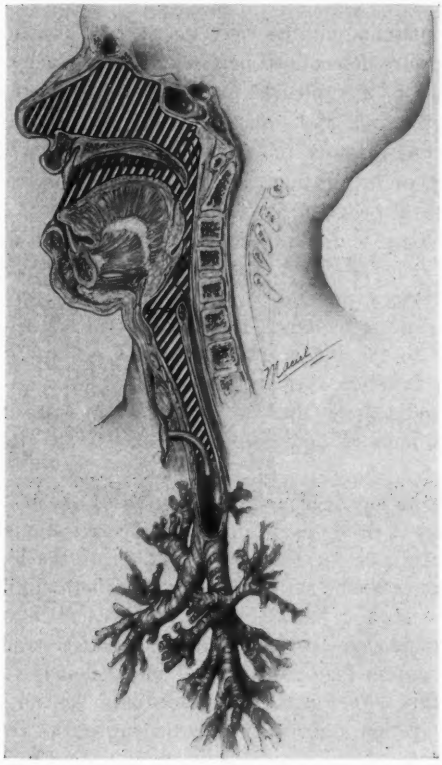

Fig. 1.—The approximate volume of the respiratory tree eliminated by tracheotomy is represented by the cross hatched area. The result is a decreased dead space and diminished resistance to the respired air.

grams were anesthetized with intravenous nembutal, a long curved incision was made over the anterolateral portion of the left chest, and ribs 2 to 8 were exposed and divided anteriorly and posteriorly so as to produce a large loose segment of the thoracic cage. The wounds were closed after mushroom catheters had been placed in each pleural cavity. Intrapleural pressures were recorded before and after tracheotomy by means of a water manometer connected with the catheters. The results are shown in Table I. Though there may be some question as to the absolute accuracy of readings made with a water manometer the consistent fall in intrapleural pressure following tracheotomy is of real significance.

TABLE I. INTRATHORACIC PRESSURE IN DOGS BEFORE AND AFTER TRACHEOTOMY

DOG NUMBER	BEFORE TRACHEOTOMY C.C. OF H_2O		AFTER TRACHEOTOMY C.C. OF H_2O	
	INSPIRATION	EXPIRATION	INSPIRATION	EXPIRATION
1	−5	+2	−3	+1
2	−5	−1	−3	−1
3	−4	+2	−2	+1
4	−4	+1	−2	−0
5	−5	−1	−3	−1

In two human patients, needles were inserted into each pleural cavity and the intrapleural pressure determined before and after tracheotomy with a water manometer. In Case 5, intrapleural pressure pre-tracheotomy varied between minus 7 cm. on inspiration and plus 4 cm. on expiration. Following tracheotomy, the intrapleural pressure fell to oscillations between minus 3 on inspiration and plus 2 cm. on expiration. In Case 6, the pre-tracheotomy readings showed the intrapleural pressure to oscillate between minus 7 and minus 2 cm., while after tracheotomy it fell to between minus 3 and minus 1 centimeter. From the limited number of observations on experimental animals and in human patients with respiratory distress, it seems evident that tracheotomy, by defunctioning the upper air passages, the larynx, and a portion of the trachea, decreases resistance to the inflow and outflow of air and thereby contributes to better pulmonary function. Again it should be emphasized that in those cases in which adequate pulmonary function is maintained with difficulty, any factor which contributes to more efficient respiration may be the determining influence in survival.

A corollary of the more efficient functioning of the respiratory system resulting from decrease in dead space and diminished resistance to inflow and outflow of the respired air is the lessened excursion of the loose segment of the thoracic wall. This has been a striking observation both clinically and in experimental animals (see Figs. 2 and 3). The paradoxical movements of the free segment of the thoracic cage are greatly decreased after tracheotomy. It is believed that this is a potent factor in survival and that it is a result of the lessening of the respiratory effort needed for adequate ventilation. In turn, this greater ease of respiration and lessened movement of the fractured ribs results in marked diminution of pain and abolition of apprehension. These factors are of real importance in contributing to better aeration.

The beneficial effects of tracheotomy in the management of respiratory embarrassment should be reflected in the oxygen saturation of the arterial blood. In Case 5, before tracheotomy and while receiving oxygen, the oxygen saturation was 85 per cent with a CO_2 content of 29.7 volumes per cent. After tracheotomy, the oxygen saturation rose to 90 per cent despite the fact that the patient was no longer receiving oxygen, and the CO_2 fell to 27.9 volumes per cent.

Whether or not our explanations of the physiological and mechanical effects of tracheotomy are proper, the fact remains that in our clinical experience it has been strikingly successful in the management of patients with *severe* crushing injuries of the thorax. Its use should be reserved for such cases, since there are

many patients with lesser degrees of injury, and even some with greater ones who maintain adequate oxygenation if relieved of pain by intercostal nerve block and if supplied with oxygen by nasal catheter.

Our observations and experience suggest that in addition to its employment in patients with crushed chest, tracheotomy be recommended on physiologic grounds in those instances in which "paradoxical movement" or low tidal volume are of serious moment. Such conditions are encountered (1) in cases

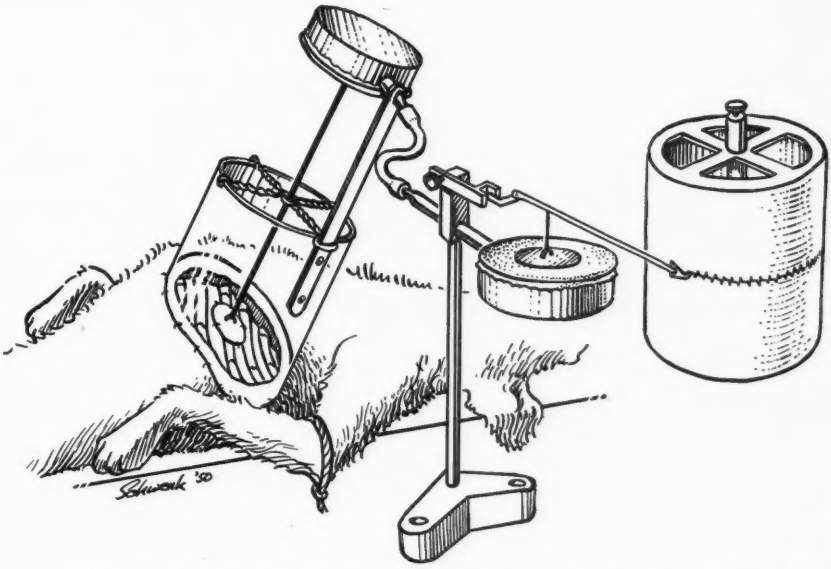

Fig. 2.—Apparatus for recording oscillations of a segment of the thoracic wall. A large, light metal cannister with a diameter exceeding that of the injured segment is sutured in place to normal thoracic wall. This serves as a support for an inner piston formed of a thin metal rod suspended as a plumb line by rubber bands and attached at either end to the moving thoracic wall and an ordinary tambour. Movements of the thoracic wall are then transcribed on a revolving drum.

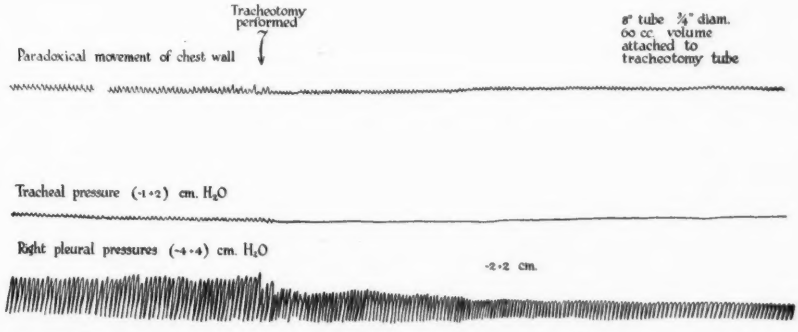
Tracheotomy performed
Paradoxical movement of chest wall
8" tube ¾" diam. 60 cc volume attached to tracheotomy tube
Tracheal pressure (-1·2) cm. H_2O
Right pleural pressures (-4·4) cm. H_2O
-2·2 cm.

Fig. 3.—Tracing made with the apparatus shown in Fig. 2 indicating the decreased paradoxical movement of the loose segment of the thoracic wall after tracheotomy. Note that the intrapleural and intratracheal pressures are likewise diminished.

in which too extensive rib resections have been performed in thoracoplasty or in the removal of large tumors of the sternum or ribs, and (2) in cases with unstable mediastinum in the presence of open pneumothorax. We have used it with great success in one case of the latter type.

CASE 7.—H. M. V. (245559). This 46-year-old white man was admitted to the Cincinnati General Hospital on Dec. 2, 1948, with advanced bronchiectasis involving the right upper and middle lobes. Lobectomy was performed on Dec. 16, 1948. He developed a bronchopleural fistula and empyema and an extensive rib resection for drainage was done on March 10, 1949. The patient was readmitted to the Cincinnati General Hospital in May, 1950. At that time he was emaciated and chronically ill from long-continued suppuration in the lung and pleural cavity. His empyema cavity measured apparently 500 c. c., but was widely exteriorized by the former extensive rib resections. The remaining right lower lobe showed evidence of marked bronchiectasis. On May 26 the right lower lobe was resected with some difficulty because extrapleural mobilization of the lung was necessary. After resection of the entire seventh rib, the hilar structures were isolated and ligated individually, and the bronchial stump was closed with interrupted mattress sutures. The entire right pleural cavity was loosely packed with a gauze roll, the end of which was brought out through the previously formed window in the thorax, and the operative wound closed. Twelve hours after operation the patient had considerable respiratory difficulty which resulted both from the to-and-fro movement of the mediastinum and from the accumulation of secretions in the bronchial tree. Cough was ineffectual, frequent, and labored. The patient was cyanotic and extremely apprehensive. As a result of these factors, he had reached the point of exhaustion and was in extremis. A tracheotomy was performed at 9:00 A.M. Following this procedure the respirations became much easier and the extreme apprehension disappeared. After several aspirations, the tracheobronchial tree remained clear. There was a striking improvement in color and in ease of respiration, and the patient dropped off to sleep. There was no further respiratory distress, cyanosis, or wheezing respirations. The patient improved steadily and soon was up and about in the ward. Unfortunately on June 8, thirteen days after operation, a massive hemorrhage occurred into the right pleural cavity from erosion of one of the large hilar vessels, and the patient expired within a few minutes.

Comment.—The effect of tracheotomy in this patient with marked respiratory distress, from accumulation of bronchial secretions and from paradoxical movements of the mediastinum as a result of a widely opened right thorax, was striking. The cyanosis and labored respirations disappeared, as did the marked apprehension and pain. In spite of repeated aspiration of the tracheobronchial tree by nasal catheter before tracheotomy, the cyanosis and respiratory difficulty had persisted. Not only did tracheotomy permit easy continued aspiration of excessive bronchial secretions, its physiologic effects were considered lifesaving in this instance.

SUMMARY

1. Favorable clinical experience with the use of tracheotomy in the management of six cases of severe crushing injuries of the thorax has been presented.

2. The beneficial effects of tracheotomy are both mechanical and physiological. The most important of the former is that the tracheobronchial tree can be easily and frequently aspirated by any member of the house or nursing staff. Of the latter, the suggested benefits are the decrease in volume of the dead space in the respiratory tree and the diminished resistance to inspired and expired air. As corollaries of these are the diminished excursion of the loose segment of the thoracic wall and the relief of pain.

3. The use of tracheotomy is advocated in cases of severe crushing injuries of the thorax and in other instances in which paradoxical movement and low tidal air are factors of importance. When indicated, it should be employed before respiratory difficulties become alarming.

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COARCTATION OF THE AORTA AT THE LEVEL OF THE DIAPHRAGM TREATED SUCCESSFULLY WITH A PRESERVED HUMAN BLOOD VESSEL GRAFT

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SURGICAL correction of coarctation of the aorta has become common since Crafoord reported his first case in 1945. Prior to the introduction of aorta grafts for replacement of long coarctations by Gross in 1948, excision of stenosed aortic segments was limited to relatively localized constrictions. Development of this technique has appreciably reduced the number of inoperable cases due to long coarctations, and in 1950 Gross reported the use of this technique in 12 of his 130 cases. Although the grafts employed by Gross were five to six centimeters in length, longer ones have been used successfully, such as the one employed by Swan in 1950 (52-day-old graft eight centimeters in length).

Schlesinger reported the first case of coarctation of the aorta at the level of the diaphragm in 1835. Of sixteen cases having constrictions distal to the locations usually encountered, eight were found in the lower thorax near the diaphragm.[1, 4, 10, 12, 15, 17]

Contemplated surgical correction of this type of anomaly has previously been unsuccessful.[1, 12] Kondo and his associates considered low coarctations theoretically curable, while Freeman and his co-workers have recently indicated that a positive diagnosis of such a defect by contrast visualization of the aorta has eliminated unnecessary exploration. Experience gained in the case in this report indicates that surgical correction may now be extended to include the coarctations located below the usual site. This is the first case of coarctation of the aorta at the level of the diaphragm successfully treated with a preserved blood vessel graft.

CASE REPORT

A 19-year-old housewife entered Walter Reed Army Hospital on June 23, 1950, with a diagnosis of coarctation of the aorta. Three years prior to admission physical examination revealed the presence of hypertension in the arms. The patient stated that she was told her blood pressure at that time was 180 millimeters of mercury systolic. In retrospect, she had noticed that if she exercised vigorously, she developed pain behind the knees and in the calves of both legs. For one and one-half years prior to admission, she became aware of increasing fatigability and slight shortness of breath. She had no other symptoms.

Three months before admission, a physical examination at an Army hospital revealed hypertension in the upper extremities, and absent pulsations in the lower extremities. No

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blood pressure could be measured in the lower extremities when ordinary clinical methods were used. Following this examination, she was referred to the Walter Reed Army Hospital.

She was seen in the Cardiology Clinic at Walter Reed Hospital where the clinical findings mentioned above were confirmed. In addition, the following findings suggested the clinical diagnosis of a lower thoracic or abdominal coarctation of the aorta. She had a systolic bruit of maximal intensity at the level of the ninth and tenth thoracic vertebrae posteriorly and in the epigastrium anteriorly, abnormal pulsations around the scapulae and posteriorly in the lowermost intercostal arteries, roentgenographic evidence of notching of the lowermost ribs with a normal appearing aortic knob (Fig. 1), and fluoroscopic demonstration of a normal aorta as far as it could be traced into the cardiac silhouette. The patient was admitted to the hospital with the diagnosis of coarctation of the aorta, probably located in the lower thoracic or abdominal aorta.

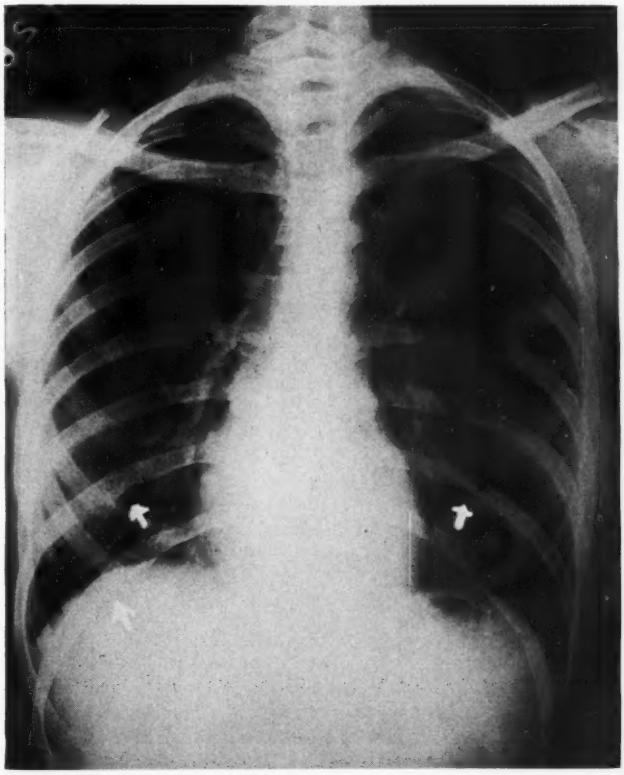

Fig. 1.—Posteroanterior roentgenogram of chest, taken preoperatively. Note normal aortic knob and rib notching most marked in the ninth and tenth ribs.

The past medical history, family history, and social history were noncontributory.

On admission to the hospital, physical examination revealed a well-developed, normally nourished, white woman in no apparent discomfort. The temperature was 98.6° F., the pulse rate was 78 beats per minute, and the respiratory rate was 20 per minute. The weight was 123 pounds, and the height was 66.5 inches. The positive physical findings were as follows: strong pulsations were present at the base of the neck bilaterally. The heart appeared to be

of normal shape to percussion. The heart rate was normal, and there were no murmurs. The aortic second sound was louder than the pulmonic second sound. Pulsations were easily palpable under the lower ribs, especially on the left side. There was a systolic bruit heard best posteriorly on the left at the level of the ninth and tenth thoracic vertebrae. The abdomen was scaphoid, and no pulsations of the abdominal aorta could be detected. Examination of the upper extremities revealed normal pulsations and a blood pressure of 180/110. No pulsations were felt in the lower extremities, nor was the blood pressure measurable with a blood pressure cuff. Oscillometric readings in the lower extremities were found to be diminished showing one-third unit of deflection.

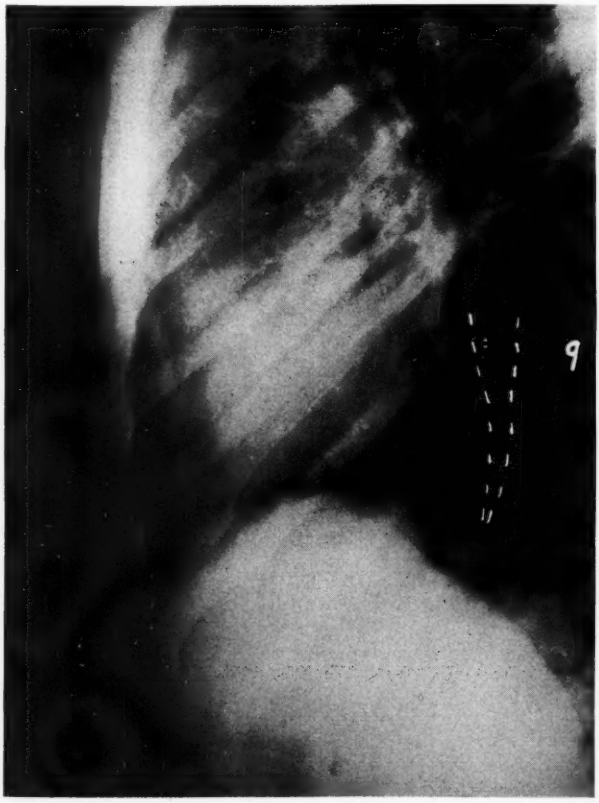

Fig. 2.—Angiocardiogram which demonstrates that the coarctation begins at the body of the ninth thoracic vertebra.

Laboratory examinations, including hemograms, serology, sedimentation rate, total and fractional serum proteins, serum chloride, blood urea nitrogen, and circulation time were all within normal limits. Phenolsulfonphthalein excretion in two hours showed 64 per cent on the first examination and 59 per cent on the second examination. Urea clearance was 85 per cent of normal. Intravenous pyelography was normal. Three angiocardiograms were made before satisfactory films were obtained. The angiocardiogram revealed a constriction of the descending aorta at the level of the ninth thoracic vertebra. (Fig. 2.)

TREATMENT

The diagnosis of lower thoracic coarctation was confirmed, and it was decided to attempt surgical correction of the lesion. The operation was performed Sept. 27, 1950, under endotracheal anesthesia. Adequate exposure was obtained by resection of the ninth left rib for practically its entire length. Exploration of the left hemithorax revealed a normal aorta down to the ninth thoracic vertebra. The mediastinal pleura was opened, and the proximal end of the stenotic segment was dissected free of the surrounding tissue. The dissection

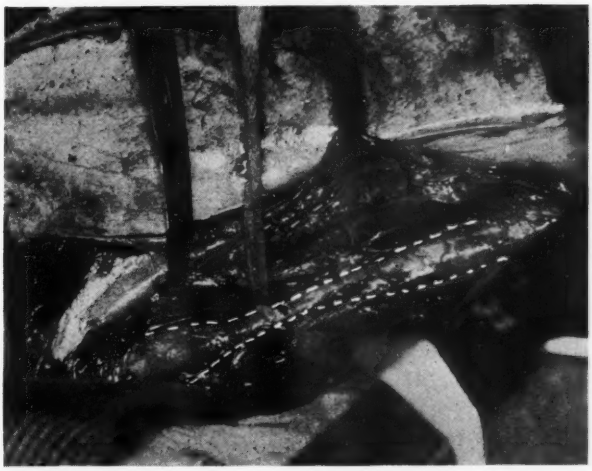

Fig. 3.—Photograph taken during the operation after the stenotic area was partially dissected. It is approximately seven centimeters from the normal aorta on either end of the graft.

Fig. 4.—Specimen photographed after removal. Scale here is in inches. This specimen measures 2.2 centimeters in length and 4 millimeters in outside diameter at the narrowest part. Additional tissue had been trimmed from either end of the aorta, but this tissue does not appear in this photograph.

was continued and the area of coarctation was followed through the diaphragm. The aortic hiatus was opened wider so that normal abdominal aorta could be reached just below the inferior diaphragmatic surface. The caliber of the aorta at the site of the coarctation was uniformly four millimeters in external diameter. The coarctation was approximately six and one-half centimeters in length. (Fig. 3.) It was decided to resect the stenosed segment and to bridge the defect with a preserved arterial graft. Arteries had been obtained from a

Fig. 5.—Photograph of graft in situ.

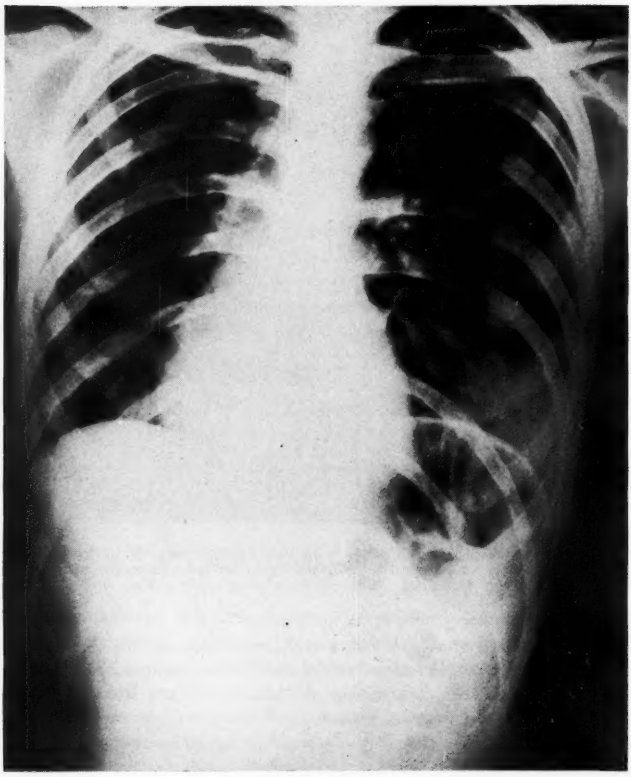

Fig. 6.—Posteroanterior roentgenogram taken on the ninth postoperative day.

40-year-old man who had died from trauma twenty-three days previously. The graft had been preserved in a modified Tyrode's solution and kept at 4° C. A piece of the iliac artery which measured seven centimeters in length and fourteen millimeters in external diameter in its completely relaxed state appeared adequate for our needs. The coarctation (Fig. 4) was excised between Potts clamps, and the graft sutured in place, using continuous 0000 silk everting sutures. The distal end of the excised segment had been cut obliquely in order to preserve all branches of the abdominal aorta. As soon as the clamps were released, there were excellent pulsations in the graft and the aorta below the graft. Pulsations were palpable in the dorsalis pedis and the posterior tibial arteries. Fig. 5 illustrates the graft in situ.

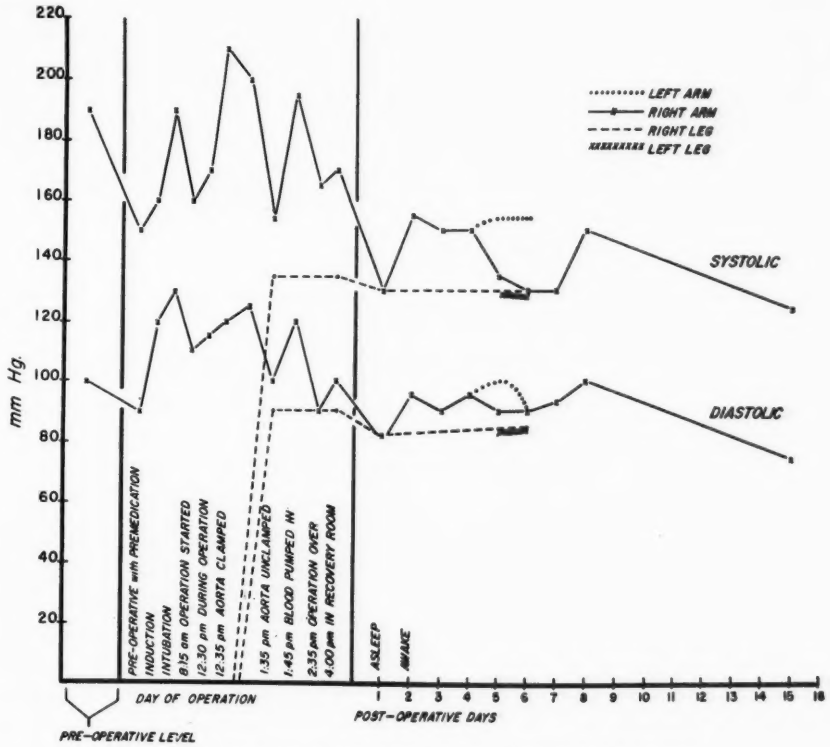

Fig. 7.—This chart depicts the changes in the patient's blood pressure.

The patient withstood the procedure well. Fifteen hundred cubic centimeters of blood were given during the operation.

Follow-up communication from the patient indicated that the blood pressure in her upper extremities has been maintained in the vicinity of 148/78. In the lower extremities the blood pressure has been approximately 122/88. The patient remarked that in general she has been feeling better than at any time for several years.

SUMMARY

This is the eighth case to be reported of coarctation of the thoracic aorta occurring at the level of the diaphragm, and the first case treated successfully with a preserved arterial graft.

The clinical features of hypertension in the arms with no palpable pulsations nor measurable blood pressure in the lower extremities combined with a systolic bruit of maximal intensity at the ninth and tenth thoracic vertebrae, fluoroscopic demonstration of a normal aorta as far as the middescending portion, and notching of the lower most ribs, suggested the diagnosis. Angiocardiography successfully demonstrated the correct location of the coarctation.

Thoracic exploration was postponed until preserved arterial grafts were available since the probability of a long stenosed segment was recognized. Adequate exposure was obtained by subperiosteal resection of the ninth rib. At operation a coarctation of the aorta six and one-half centimeters long was found which extended from the ninth thoracic vertebra to the first portion of the abdominal aorta. The stenosis was dissected free, excised, and replaced by a 23-day-old graft seven centimeters in length. A piece of the blood vessel used as a graft grew in tissue culture when it was thirty-nine days old. Postoperatively the patient convalesced uneventfully, and at the present time has relatively normal blood pressures in her extremities. (Figs. 6 and 7.)

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AN EFFECTIVE METHOD FOR BRONCHIAL CLOSURE

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ALTHOUGH bronchial closure following pulmonary resection has been fairly well standardized and the incidence of reopening has been greatly diminished, this unfortunate complication still occasionally occurs. A simple modification of the accepted methods of covering the stump with pleura has proved to be so successful in the past year that it is thought to be worth while briefly to present the technique used.

The experience with this technique of bronchial closure is too limited to be of statistical significance, but from a practical standpoint it seems well worth reporting. It has been used in thirty-five cases of pulmonary resections. These were lobectomy, segmental resection, and pneumonectomy for a variety of conditions including neoplasms and infections, both nontuberculous and tuberculous. There was no evidence of bronchial disruption in any case, but in one there was evidence of leak. In the latter a leak was assumed because of the development of an empyema with some production of bloody mucus several weeks following a left lower lobectomy and lingulectomy for bronchiectasis. The empyema was drained openly and satisfactory healing occurred without evidence of persistence of a fistula.

Rienhoff and associates[1] in 1942 reviewed the subject of bronchial closure and drew important conclusions based on experimental and clinical observations. They stressed the important factors concerned with the healing of the severed bronchus. These were that the cut bronchus heals at its end, the importance of preserving a viable stump by avoiding trauma to it or to any of its vessels, and the importance of covering the cut end of the bronchus with adjacent mediastinal pleura. They obtained healing by several methods: one of which was simply to leave the bronchus widely open with pleura sutured against it; another was to place the suture line as far proximal as possible from the cut end, covering the latter with pleura only. Their most frequent and serious complication was opening of the bronchial stump with the development of a fistula.

In 1945 Sweet[2] presented a method of bronchial closure based on the above principles and stressed, among other things, the importance of covering the cut end of the bronchus with pleura by suturing the edge of a pleural flap to the peribronchial tissue proximal to the sutured end.

A method of bronchial closure which, although it may not be original, has proved entirely satisfactory during the past year, is presented.

The method used is as follows: the part to be resected is first mobilized down to its bronchus by dissection and division of its vascular supply and peri-

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bronchial attachments, except that in segmental resection the vessels and bronchus may be isolated first. The bronchus is then clamped close to the lung tissue distal to the point of planned section to prevent any spillage from the diseased portion. Sutures of No. 0000 silk on small straight needles are then passed through the bronchus about 3 mm. proximal to the line of section. This entire line of marginal sutures is placed, keeping each suture in order. Gauze is then placed around the bronchus to guard against any possible contamination.

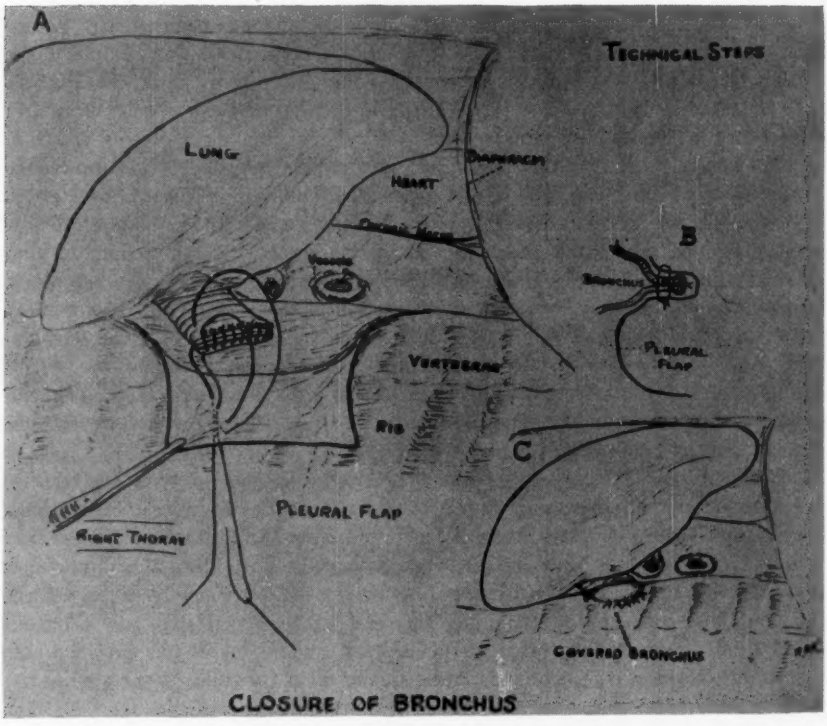

Fig. 1.—Steps in closure. *A*, First vertical mattress suture in place proximal to marginal row of bronchial sutures; others to follow in order. *B*, Section of bronchus showing sutures in place and tied. *C*, Completed closure.

These sutures are then held aside while the bronchus is cut across directly with the knife, or with a right angle scissors, and the sutures are then held up snugly as a unit. The bronchus is thus open a very short time. The sutures are then tied snugly, one by one, approximating the membranous portion to the cartilaginous portion of the bronchus. One or two additional sutures may then be placed if necessary for an airtight closure. A variation of this part of the procedure is to place and tie each suture as the bronchus is cut across as described by Sweet (1945). The straight needle is believed to be better than a curved because the point is more easily and definitely directed.

The marginal row of sutures is used to manipulate and to hold the bronchus while it is covered with adjacent pleura, this being elevated as a flap, or simply shifted, usually from the posterior vertebral area. In Fig. 1, *A* several vertical mattress sutures of fine silk are placed through the bronchus just proximal to the marginal row of sutures, each of these sutures anchoring the pleura snugly over the bronchus both anteriorly and posteriorly in glovelike fashion. The nonserous surface of the pleura is brought closely against the cut end of the bronchus. Each suture is passed through an edge of the pleura, then through the bronchus, and through the other edge of the pleural flap. It is then brought directly back alongside itself through pleura, bronchus, and pleura close to the starting point. The pleura is brought nicely over the entire cut end of the bronchus as these sutures are tied after all have been placed (Fig. 1, *B* and *C*).

This method of closure approximates the bronchial mucosa proximal to the cut end of the bronchus and the marginal sutures. Though this may be a disadvantage, it has not thus far proved to be so. The method does tend to bring the subpleural areolar tissue in close apposition to the end of the bronchus without leaving appreciable, actual, or potential space between it and the pleura. This should further insure good healing.

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A BRONCHIAL YARDSTICK

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WITH the development of thoracic surgery, in recent years, a need for an easy and accurate means of obtaining linear measurements within the tracheobronchial tree has been increasingly apparent. For an example, one of the things a chest surgeon needs to know is how much distance there is between a bronchial lesion and the carina. For the purpose of surgery a report from the endoscopist to the effect that it is so many centimeters from the incisor teeth, or an estimate of two to three centimeters beyond the carina is of little value. The thoracic surgeon has been justified in his criticism of such reporting, and it has forced him, in many instances, into doing his own bronchoscopy when he is already carrying a too heavy work load.

With the hope of starting endobronchial studies on a more satisfactory basis from the point of view of accurate quantitative examination the Bronchial Yardstick has been designed. It allows the endoscopist quickly, easily, and safely to obtain the necessary measurements without undo prolongation of the diagnostic bronchoscopy.

The instrument consists of a brass rod (or tube) 1.5 mm. in diameter and 60 cm. in length. On this rod are mounted, in unattached tandem, two jewelers' pin clutches of diameter suitable to the lumen of the bronchoscope to be used, so vision by them is possible. These pin clutches are mounted in direction such that they are locked in the direction of the peripheral end of the rod, and yet will move easily up the rod, thus eliminating danger of their dropping off the end of the rod into the tracheobronchial tree.

The proximal pin clutch is fitted with a transverse spike so that as the instrument is carried into the bronchoscope the clutch is automatically carried up the rod and away from the peripheral clutch. A small fitting at the end of the rod serves two purposes: (1) to prevent possible escape of a faulty pin clutch, and (2) it acts as a sort of gun site making it easier to accurately place the end of the rod at the desired spot, i.e., the border of the lesion in question.

The technique of bronchial linear measurements with the above described instrument is easy, and to the experienced endoscopist the diagramatic illustration (Fig. 2) will be almost self-explanatory. The two pin clutches are set at the point desired, the rod is then advanced into the trachea or bronchus to the further extremity of the line between the two points in question. The instrument is then withdrawn and the distance between the two pin clutches is measured in millimeters. Thus, it is possible, by successive settings of the clutches, not only to measure the distance from the carina to the lesion, but also the size of the lesion, in respect to the length of the bronchus, may also be measured.

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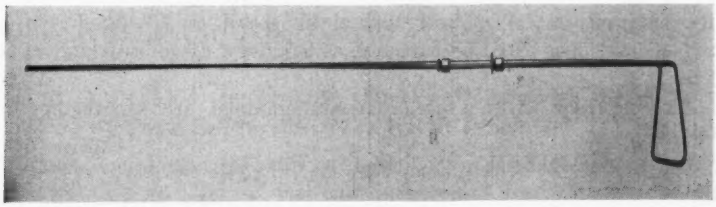

Fig. 1.

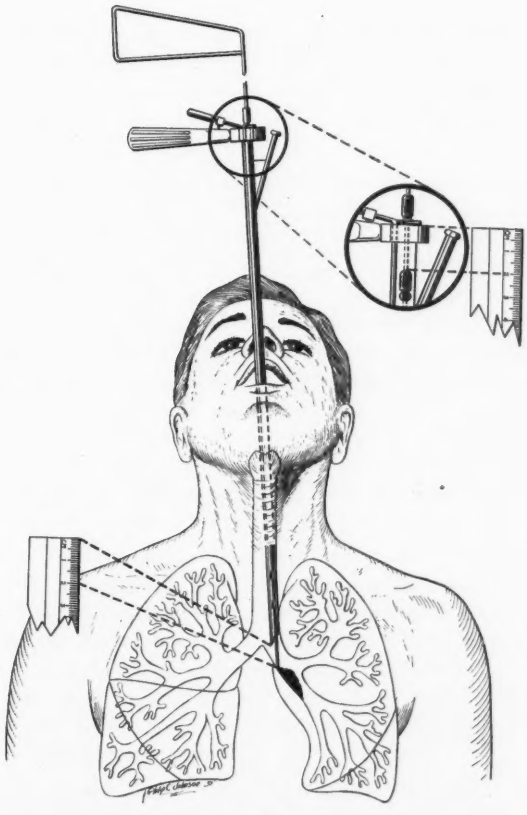

Fig. 2.

SUMMARY

1. An instrument, termed a Bronchial Yardstick,* for obtaining linear measurements within the tracheobronchial tree has been described.

2. The instrument has been found satisfactory as to ease and safety in actual use.

3. The accuracy of the measurements obtained are satisfactory to the thoracic surgeons.

4. It is hoped that the introduction of this instrument may mark the beginning of more accurate endoscopic reporting.

*The Bronchial Yardstick may be obtained at The Foregger Company, Inc., 55 West 42nd Street, New York 18, N. Y.

CLINICALLY INDETERMINATE PULMONARY FIBROSIS
A PATHOLOGIC STUDY*

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IN THE study of pulmonary disease the physician is often confronted with the problem of diagnosis and treatment of a patient who has shortness of breath, cough, and at times cyanosis, either with or without positive physical signs. The roentgenogram of the thorax shows bilateral parenchymal infiltration of indeterminate nature, and additional study may fail to reveal the underlying cause. The term "pulmonary fibrosis" is often used to categorize these patients.

Mallory[1] defined pulmonary fibrosis as "A condition characterized by multifocal or generalized fairly evenly disseminated pulmonary lesions, the origin of which is dubious." Such a definition is supported clinically by the roentgenogram of the thorax, which presents the only uniformly objective evidence of pulmonary disease.

It is only by adequate pathologic study that any type of classification can be made. Mallory,[1] in a review of the records of 6,000 necropsies, found the anatomic diagnosis of pulmonary fibrosis in fifty-nine instances, in only twenty-three of which the patient had had any pulmonary symptoms. He divided these cases into four groups, as follows: (1) organization of intra-alveolar fibrinous exudate, as seen in bronchitis or pneumonic exudation without resolution; (2) healed infarction; (3) healed destructive pneumonitis and infections producing necrosis, as seen in infectious granulomas, and septic mixed infections which are usually associated with abscess formation or bronchiectasis; and (4) interstitial fibrosis, either in alveolar walls or lymphatic channels. In this group are mentioned prolonged chronic passive congestion from valvular cardiac disease, interstitial pneumonia, sarcoid, viral or rickettsial infections, chemical irritation, as well as still other processes.

Some of the characteristics of the lung, said to have fibrosis, and regardless of the specific etiologic agent, are increase in the collagenous tissue, and destruction and distortion of the elastic elements with loss of the normal recoil power of the parenchyma. The alveoli are obliterated which in turn decreases the area of ventilating surface. The scarred walls have decreased vascularity in the end stages, which produces diminished aeration of blood. Metaplasia of the alveolar lining cell to a cuboidal type may occur, leading to an alteration in function of the alveolar lining.

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In 1932, Cooper[2] tabulated twelve conditions which must be considered in the differential diagnosis of bilateral, fairly evenly disseminated lesions. Austrian and Brown,[3] in 1942, listed twenty-five causes for such a roentgenographic picture, and by 1948, Nelson and Heublein[4] mentioned seventy-five different causes. An etiologic classification of pulmonary fibrosis, as modified from a summary by King,[5] is presented in Table I.

In an attempt to clarify the diagnostic term "pulmonary fibrosis," we made a study of cases in which a clinical diagnosis of bilateral diffuse pulmonary fibrosis of indeterminate origin had been made and in which necropsy specimens were available. Details of the study follow.

SELECTION OF CASES

A review was made of the cases at the Mayo Clinic from Jan. 1, 1920, through Feb. 28, 1950, in which a clinical diagnosis of bilateral pulmonary fibrosis or indeterminate bilateral pulmonary infiltration was made, and in which the roentgenograms of the thorax revealed disseminated lesions. All cases in which the etiologic agent was known or strongly suspected were excluded from this series, as were those in which the data was insufficient for adequate study. No cases in which an occupational hazard might have contributed to the "fibrosis" were included. Diseases such as Boeck's sarcoid, asthma, senile emphysema, proved or probable bronchiectasis, chronic bronchitis, tuberculosis, or other proved parenchymal infections were not included. Those cases of fibrosis associated with rare systemic diseases such as scleroderma or xanthomatosis were excluded. The only other criterion for selection was whether or not necropsy had been performed. Most of the instances of indeterminate diagnosis could not be included in the detailed study, because of lack of postmortem examination, or because necropsy had been done elsewhere and the findings were not known to us. An attempt was made to select only those cases in which the clinician had been unable to make or suggest a definite diagnosis.

TABLE I. ETIOLOGIC CLASSIFICATION OF PULMONARY FIBROSIS

Infectious diseases	Circulatory disturbances
Inhalation diseases	Skeletal abnormalities
Neoplastic diseases	Postradiation changes
Diseases of blood	Emphysema, asthma, asthmatic bronchitis
Systemic diseases	Indeterminate
Bronchiectasis	Indeterminate

The cases records of 647 patients were reviewed. For fifteen of these patients there were adequate clinical data and necropsy material, and the cases are reported herein.

REPORT OF CASES

CASES 1, 2, and 3.—*Infectious Diseases:* Two patients had pathologic evidence of organized pneumonia. One was a 51-year-old woman post-office worker (Case 1), and the other a 41-year-old salesman (Case 2), both with symptoms of progressive dyspnea, and slight cough for four years and fourteen months, respectively. The roentgenograms of the chest are shown in Figs. 1*a* and 1*c*, respectively.

Microscopically, in each case there were marked replacement of alveolar exudate by fibrous tissue and obliteration of alveolar spaces (Fig. 1*b*). Alveolar and bronchial epithelial hyperplasia, alveolar hyaline membrane (Fig. 1*d*), and generalized, moderately severe vascular change consisting of thickening of the wall of pulmonary arterioles, with reduction in the size of the lumen, were other findings.

These patients suffered from gradual replacement of the functioning pulmonary bed by scar tissue. There was no history of an acute infectious process in Case 1, and the disease was not febrile until the last few days of life. There had been no known exposure to

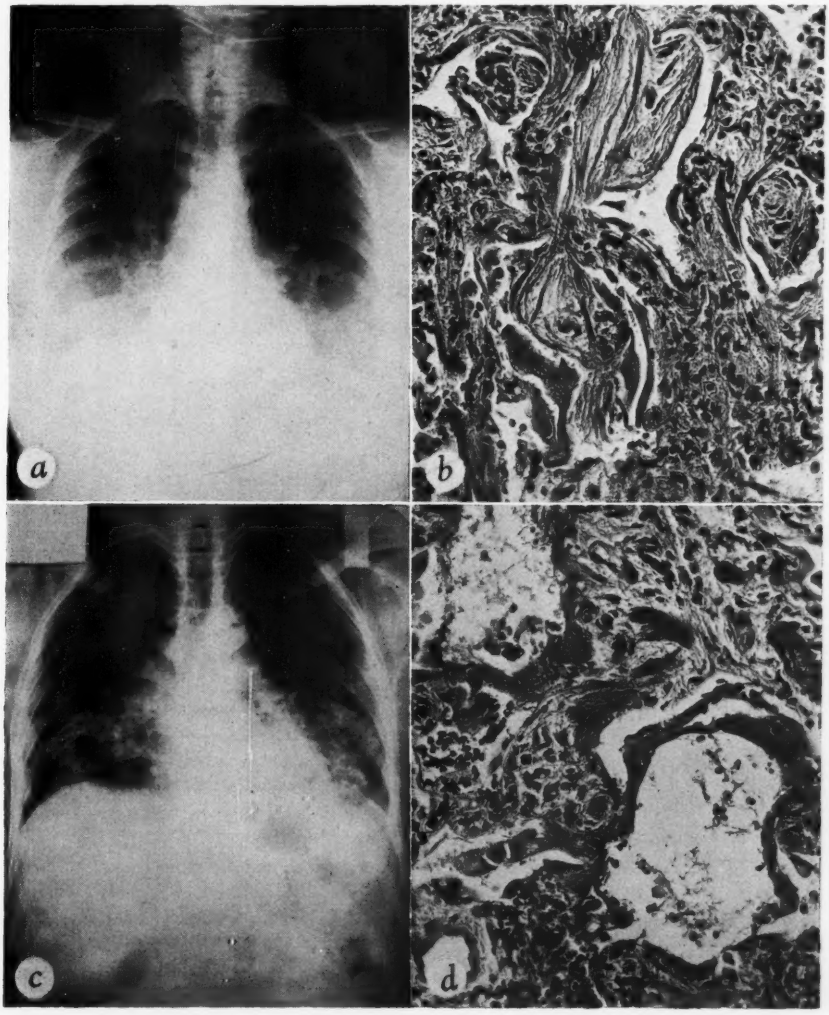

Fig. 1.—Organized pneumonia. *a* (Case 1). Roentgenographic appearance. *b*, *c*, and *d* (Case 2). *b*, Pores of Cohn and sheaves of Newman are shown (hematoxylin and eosin × 195). *c*, Roentgenographic appearance. *d*, Alveolar hyaline membrane (hematoxylin and eosin × 195).

irritants. No pathogenic organism was recovered during life, and at postmortem examination no bacteria were found in the lesions. The pathologic picture was one of an organized pneumonia. In the second case there was a history compatible with a bronchopneumonia at the onset of the illness, which is in contrast to Case 1.

The third case of infection was that of a 34-year-old salesman with a two-year history of cough, progressive dyspnea, weakness, and loss of weight (Case 3). Necropsy revealed extensive pulmonary scarring, calcification, and necrosis. All sections contained a fungus (Fig. 2). Cultures of the lungs yielded *Aspergillus glaucus*.*

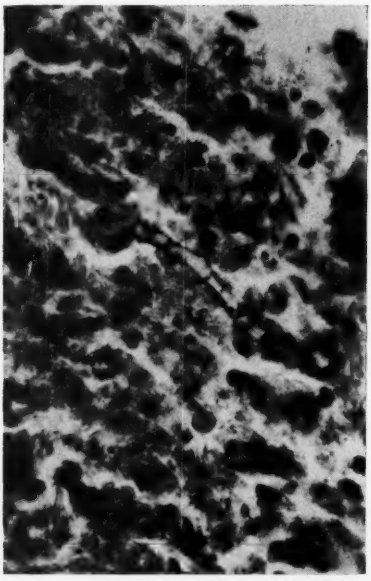

Fig. 2.—(Case 3). Fungus (Aspergillus glaucus) in pulmonary exudate (hematoxylin and eosin × 850).

CASES 4, 5, 6, and 7.—*Neoplastic Disease:* There were four cases in which neoplastic disease was found at necropsy. A 28-year-old man (Case 4) had respiratory symptoms for two months, and vague abdominal pain for one year. The roentgenogram of the chest is shown in Fig. 3*a*. He died of respiratory failure. A primary adenocarcinoma of the transverse colon was found at postmortem examination. Microscopic sections showed tumor cells distending lymphatic vessels throughout all pulmonary lobes (Fig. 3*b*). There was very little fibroblastic proliferation to explain the clinical impression of pulmonary fibrosis.

A 63-year-old farmer (Case 5) had respiratory complaints for three months (Fig. 3*c*) and vague abdominal distress for six months. The pathologic findings included carcinoma of the tail of the pancreas with metastasis to the lungs (Fig. 3*d*), liver, and right adrenal. In this case also there was no fibroblastic proliferation in the parenchyma.

A 43-year-old housewife (Case 6) had incapacitating dyspnea for nearly three months. The roentgenogram of the thorax (Fig. 4*a*) showed a bilateral indeterminate process. Postmortem examination revealed a primary carcinoma of the stomach with lymphatic metastasis

*We are indebted to Dr. J. L. Marshall of Aspinwall Pa., for the report of necropsy and the loan of microscopic sections.

to the lungs. There was marked replacement of parenchyma by metastatic carcinoma with fibrous stroma (Fig. 4*b*), in contrast to the previous cases.

A 33-year-old man (Case 7) complained of cough, shortness of breath, and fever for one year. Pathologic examination showed a lymphoma of the Hodgkin's type involving the lymph nodes, liver, lungs (Fig. 4*c*), spleen, adrenals, and bone marrow.

CASE 8.—*Disease of the Blood:* A 62-year-old farmer had symptoms for only five weeks. The peripheral blood picture was compatible with a diagnosis of aplastic anemia, while the roentgenogram of the thorax revealed a diffuse bilateral infiltration in the lungs (Fig. 5*a*).

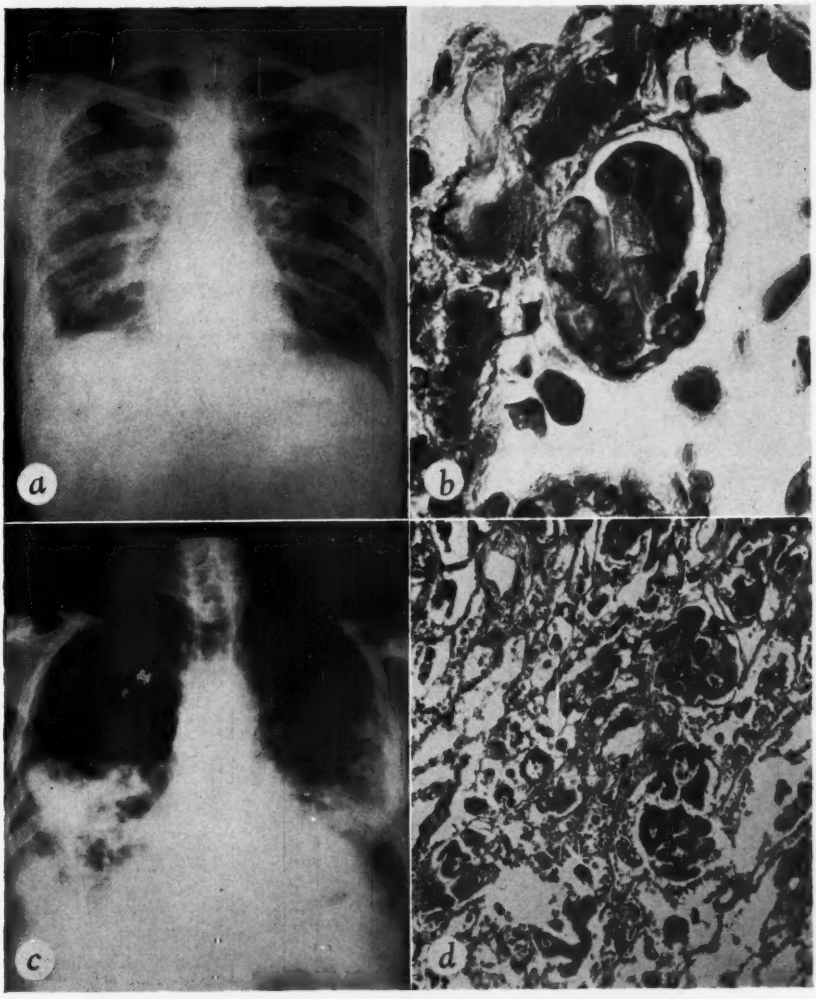

Fig. 3.—Metastatic malignant lesions in the lung. *a* and *b* (Case 4). *a*, Roentgenographic appearance. *b*, Metastatic carcinoma in a small lymphatic vessel of lung (hematoxylin and eosin × 760). *c* and *d* (Case 5). *c*, Roentgenographic appearance. *d*, Tumor cells in pulmonary alveoli (Mucin × 55).

Microscopically there were multiple focal areas of lymphocytes scattered in the alveolar walls throughout both lower lobes (Fig. 5*b*).

CASES 9, 10, 11, and 12.—*Bronchicctasis:* On pathologic examination in four cases there were extensive bronchiectasis, chronic bronchitis, patchy organized pneumonia, and varying degrees of emphysema and fibrosis (Figs. 5*d* and 6*b*). There were three women and one man, whose ages ranged from 54 to 68 years. Symptoms of cough, expectoration, and dyspnea

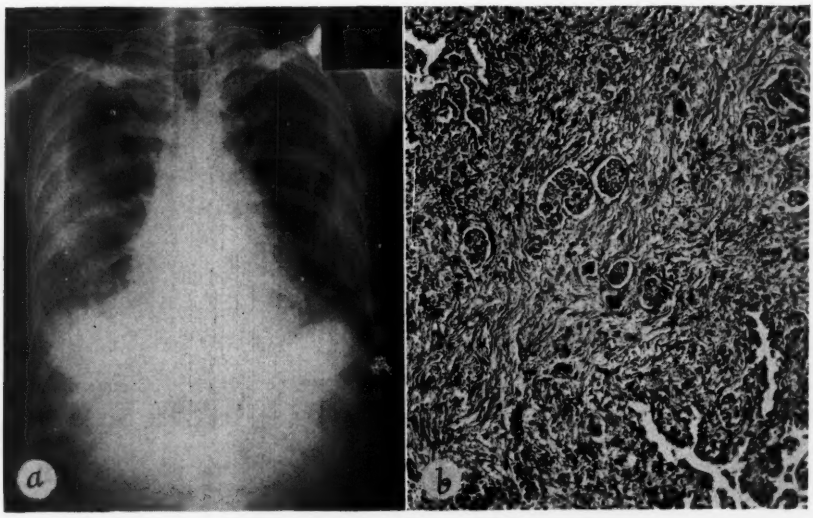

Fig. 4.—Metastatic malignant disease of the lung. *a* and *b* (Case 6). *a*, Roentgenographic appearance. *b*, Metastatic carcinoma with fibrous stroma (hematoxylin and eosin × 100).

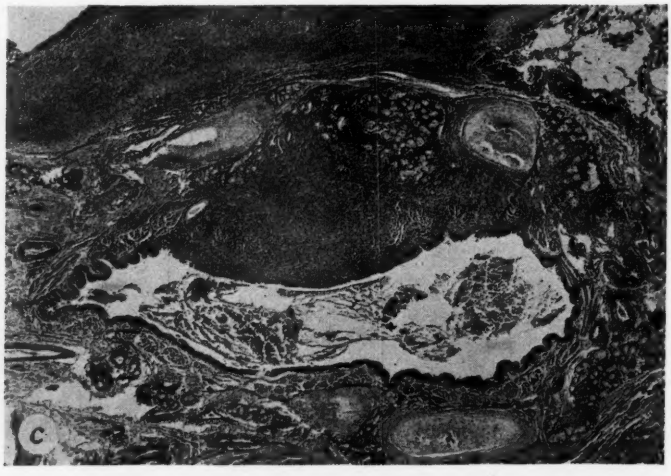

Fig. 4.—*c* (Case 7), Invasion of the bronchial wall by lymphoma of the Hodgkin's type (hematoxylin and eosin × 14).

had varied in duration from one to six years. Roentgenograms of the thorax are shown in figures 5*c*, 6*a*, and 6*c*. The most prominent symptom in this group of patients had been severe progressive dyspnea, even to the point of respiratory failure.

It is of interest that none of these patients gave a history characteristic of bronchiectasis, and all were in an older age group. It is presumed that the pathologic findings were on the basis of chronic low-grade infection producing severe destruction of pulmonary tissue and replacement fibrosis.

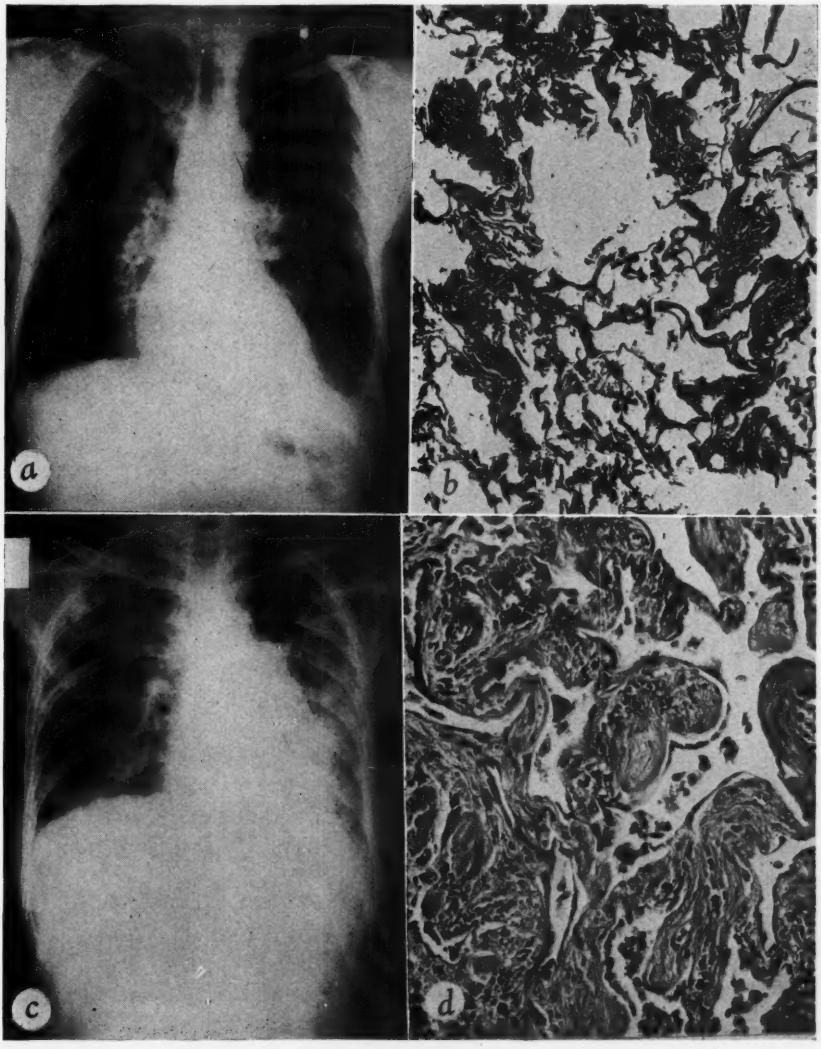

Fig. 5.—*a* and *b* (Case 8). *a*, Roentgenogram of the thorax. *b*, Focal areas of lymphocytes within the pulmonary parenchyma (hematoxylin and eosin × 43). *c* and *d* (Case 9). Bronchiectasis. *c*, Roentgenographic appearance. *d*, Bronchiectasis, parenchymal fibrosis, and cuboidal changes of the alveolar epithelium (hematoxylin and eosin × 150).

CASES 13 and 14.—*Circulatory Disturbance:* One patient in this group presented anatomic evidence of rheumatic heart disease with mitral stenosis (Case 13). This patient was a 24-year-old woman student who had symptoms of progressive dyspnea and cough for one year. The results of physical examination of the heart were negative except for tachycardia, and there were basal pulmonary râles. A roentgenogram of the thorax (Fig. 7*a*) showed a diffuse infiltration in both lower pulmonary fields and pleural effusion. Microscopically the lungs revealed a severe degree of chronic passive congestion with marked thickening of the alveolar walls, dilated capillaries, and increased number of fibroblasts and reticulum fibers

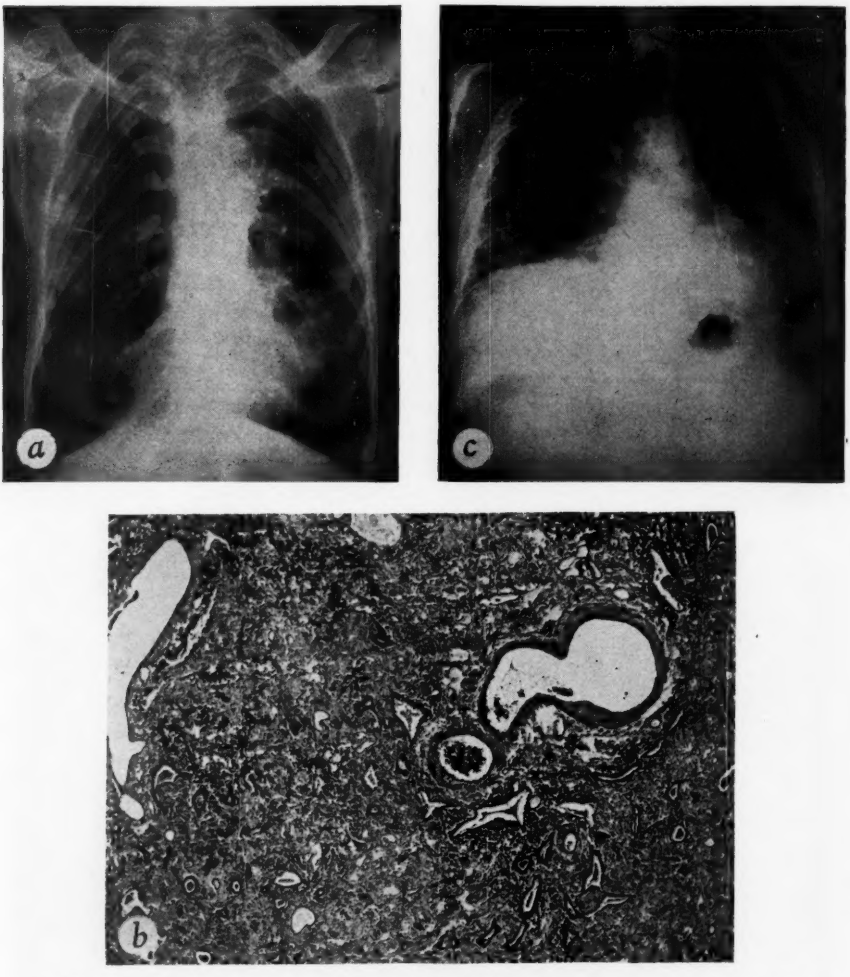

Fig. 6.—Bronchiectasis. *a* and *b* (Case 11). *a*, Roentgenographic appearance. *b*, Bronchiectasis and marked scarring of the pulmonary parenchyma (hematoxylin and eosin × 65). *c* (Case 12), Roentgenographic appearance.

(Fig. 7*b*). There was a moderately severe degree of vascular change, the muscular arteries showing thickening of the media and narrowing of the lumen. The pulmonary capillaries were dilated and tortuous.

A 54-year-old carpenter (Case 14) presented respiratory symptoms of two months' duration. The roentgenogram of the thorax (Fig. 7*c*) revealed a bilateral diffuse process

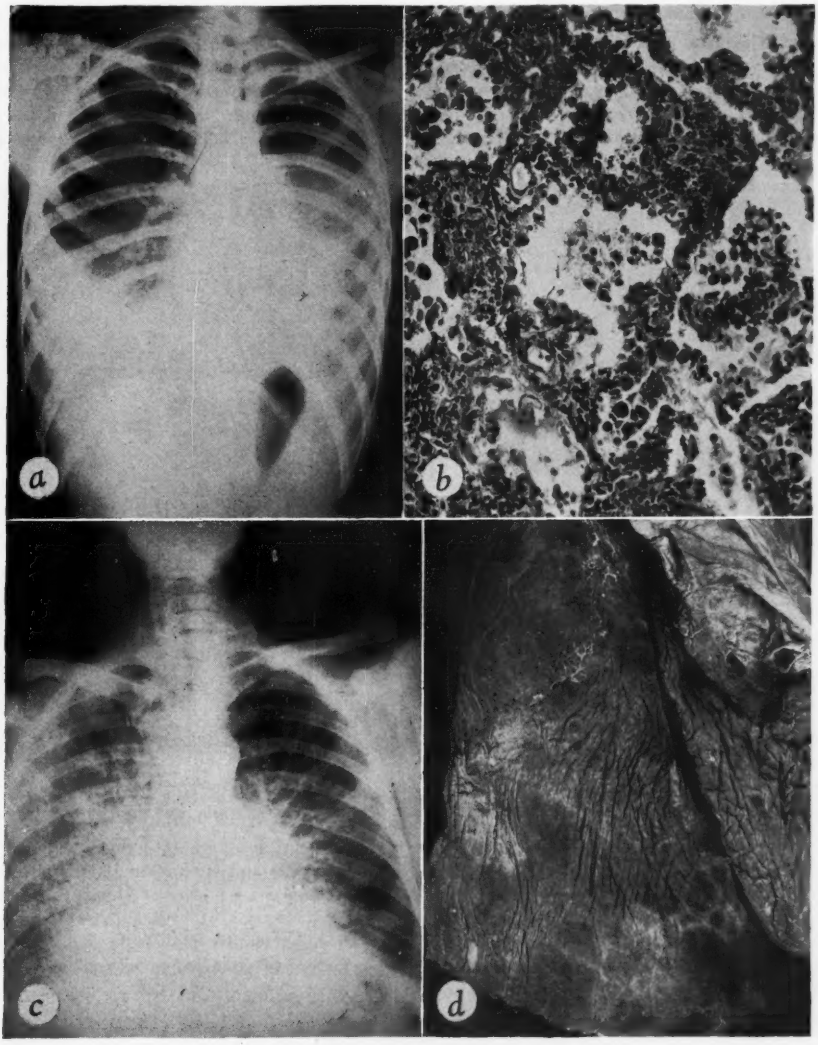

Fig. 7.—Circulatory disturbances. *a* and *b* (Case 13). *a*, Roentgenographic appearance. *b*, Alveolar-wall thickening and passive congestion secondary to mitral stenosis (hematoxylin and eosin × 150). *c* and *d* (Case 14). *c*, Roentgenographic appearance. *d*, Multiple pulmonary infarcts.

thought to represent pulmonary fibrosis. At necropsy there was periarteritis nodosa with multiple small infarcts in both lungs (Fig. 7*d*).

CASE 15.—*Indeterminate Cause:* A 47-year-old housewife had severe progressive dyspnea over a two-year period accompanied by cyanosis and clubbing of the fingers. On microscopic examination of the lungs the number of functioning alveoli was less than one-half of normal, with marked thickening of the alveolar walls (Fig. 8*a*).

This case presented features similar to those reported in four cases by Hamman and Rich,[6] in 1944, and termed "acute diffuse interstitial fibrosis." The duration of symptoms in their cases was six months or less. However, more recently other workers[7-10] have reported cases with symptoms up to six years. Pathologic features in our case, as in those mentioned by Hamman and Rich, were thickening of the alveolar walls by interstitial edema,

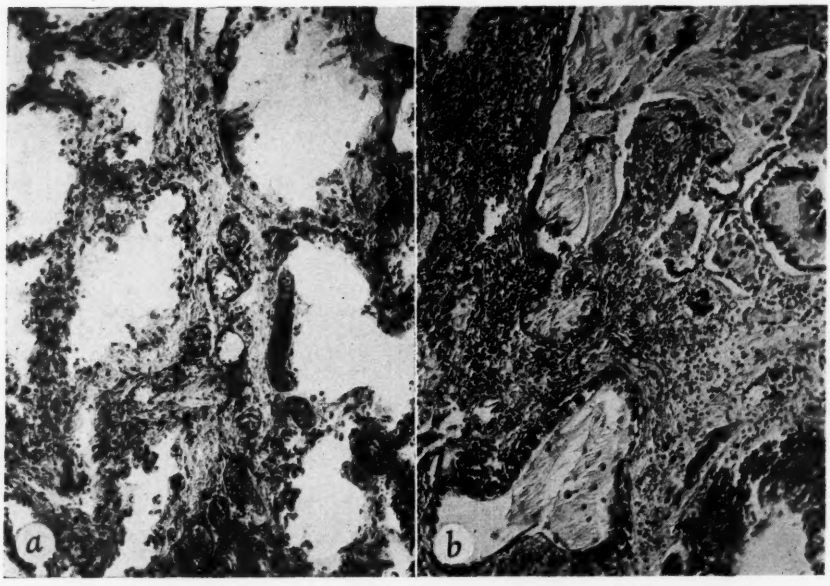

Fig. 8.—(Case 15). *a*, Vascularization and fibrosis causing thickened alveolar walls (hematoxylin and eosin × 100). *b*, Diffuse interstitial fibrosis of the lung. Distortion of the pulmonary pattern by scarring and thickening of the alveolar walls and metaplasia of alveolar lining cells (hematoxylin and eosin × 100).

increased number of capillaries, the presence of plasma cells and eosinophils, and proliferation of fibrous tissue (Fig. 8*b*). Enlargement and metaplasia of alveolar lining cells, the presence of alveolar hyaline membrane, necrosis and desquamation of bronchiolar epithelium, and focal organization of intra-alveolar exudate were other similar pathologic findings. It is felt that this case represents a distinct anatomic picture of unknown cause, and may well fit into the category "diffuse interstitial fibrosis."

COMMENT

Inasmuch as the clinical picture of "pulmonary fibrosis" may be represented by a variety of pathologic conditions, it is difficult to draw any conclusions regarding etiologic factors. As has been shown, almost any type of infection, lymphatic spread of carcinoma, and a host of unrelated diseases can

produce an indeterminate type of pulmonary process which is indeed difficult to diagnose clinically, as to etiology or pathologic anatomy.

Clinical appraisal is made by a combination of history, physical examination, and laboratory studies. The value of a complete and detailed history in pulmonary disease is obvious because many cases may be traced to occupational hazard. Wood[11] cites a typical example of such obscure etiology in his report concerning a young man who, after detailed questioning, reported that his work in an automobile factory involved exposure to silica dust in a fine oil spray.

The fifteen patients in our series had similar symptoms, although the duration of disease varied. Respiratory symptoms of less than six months' duration were seen in the cases of carcinoma, whereas the duration of symptoms in bronchiectasis and organized pneumonia was over a year. Cough, hemoptysis, and pain in the thorax were so variable that little help might be obtained from these in differential diagnosis.

Physical examination also gave few clues to the pathologic process. Auscultation of the lungs usually revealed bilateral basal râles with or without other findings. Goodrich and Johnson[12] have summarized the findings in ninety-nine cases in which cough, moist râles at the pulmonary bases, and roentgen-ray evidence of bilateral pulmonary infiltration were presented. On a purely clinical basis they could explain the condition in all but eighteen of these cases, in eleven of which there was progressive disabling shortness of breath. They concluded that a failure of the self-cleansing mechanism of the lung stimulates a fibroblastic proliferation, whether the substance retained is congestive (as in chronic valvular heart disease), infectious, metabolic, or inhaled particles. One might add malignancy to this group as has been pointed out in Cases 6 and 7, in the present series.

Clubbing of the fingers and cyanosis were usually seen in those cases in which there were anatomic evidence of right heart strain and the vascular changes in pulmonary hypertension (Cases 1, 2, 11, and 15). In Cases 9 and 13, also, there were marked pulmonary vascular changes. Hemoptysis was noted in cases of bronchiectasis and in 1 case of metastatic carcinoma.

A most valuable source of assistance comes from the laboratory study of sputum. Pathologists have pointed out the value of cytologic examination of the sputum and bronchial secretions in the diagnosis of bronchial carcinoma. In a series of 147 cases of proved or strongly suspected carcinoma of the lung studied by McDonald and Woolner,[13] the examination disclosed malignant cells in 100 (68 per cent). Although in none of the cases in our series were the results of examination positive, it is possible that more extensive cytologic study might have contributed to the correct diagnosis in Case 4. In that case, one sputum examination revealed a suspicious cell and gave a definite indication for additional procedures. In Case 5 the patient had hemoptysis which suggests bronchiolar or alveolar-wall involvement and erosion. The microscopic section in this case showed nodular areas of metastatic carcinoma replacing alveolar walls, and tumor cells in alveolar spaces (Fig. 3*d*). One might expect positive

results on sputum examination in view of this erosion, but the patient had been examined prior to employment of cytologic examination for diagnostic purposes.

The pathologist should also be given opportunity to study lymph nodes, or bronchoscopic specimens, when available, because such an indeterminate pulmonary lesion may be diagnosed by adequate examination of body secretions or biopsy. In two cases of metastatic carcinoma in this series (Cases 4 and 6), there was bilateral hydrothorax with miliary tumor nodules on both visceral and parietal surfaces. It seems reasonable to suspect that examination of the fluid from thoracic paracentesis might have given positive results for malignant cells.

The bacteriologist has a great opportunity to assist in the diagnosis of indeterminate pulmonary disease, because infection, including tuberculosis, is a major cause. In some of the cases in this series a more intensive bacteriologic study should undoubtedly have been made.

Much has been written on roentgenographic differential diagnosis in pulmonary fibrosis. It is not pertinent to this discussion to elaborate on various roentgenographic characteristics, except to mention that similarity may be noted among a variety of conditions. Robbins[14] has stated that the roentgenograms may look the same in a large group of idiopathic pulmonary fibroses. He stated, "It seems unlikely, after reviewing the known cases of fibrosis, that any distinct entity without an etiologic factor exists; but rather that other, as yet unrecognized, causes must still be sought." As has been previously mentioned, the causes given for pulmonary fibrosis have grown in number from twelve (Cooper,[2] 1932) to seventy-five (Nelson,[4] 1948).

There are still many cases in which the specific etiologic factor producing the anatomic findings is not known; however, one has only to realize the progress in the study of occupational diseases and the pneumoconioses to be encouraged. These diseases were formerly "indeterminate fibroses." In this present study it has been shown that pulmonary fibrosis is not always secondary to infection as was emphasized by Miller[15] twenty years ago. The opinion now is that many etiologic agents may provoke a fibroblastic proliferation in the pulmonary tissue. The self-cleansing mechanism, transportation of oxygen across the alveolar membrane, and removal of end products from the blood stream is thus upset by retained substances, whether infectious, metabolic, or inhaled. Metastatic carcinomas may have an inherent characteristic of provoking fibroblastic proliferation.

Any or all of the following mechanisms have a place in the pathogenesis of pulmonary fibrosis: (1) decreased or absent ciliary action, (2) impairment of bronchiolar movements, (3) occlusion of air passages, (4) plugging of lymphatics by tumor cells or cells of an inflammatory reaction, (5) failure of resolution of peribronchiolar, alveolar, or interstitial exudate, and consequent organization by fibrous tissue, (6) metaplasia of alveolar or bronchial lining cells, with consequent diminished or absent function, (7) decreased or inefficient functional respiratory volume, and (8) impairment of vascular supply.

SUMMARY

A study was made of the records of 647 cases of bilateral pulmonary fibrosis encountered at the Mayo Clinic during the period Jan. 1, 1920, through Feb. 28, 1950. Fifteen cases were selected for detailed study on the basis of the following criteria: (1) clinical diagnosis of bilateral pulmonary fibrosis of unknown cause, (2) adequate case history, physical examination, and laboratory study, and (3) pathologic specimens available. A brief account of the clinical picture and postmortem findings in each of these fifteen cases has been presented.

In the fifteen cases the following pathologic conditions were observed: infectious disease, three times; malignant neoplastic disease, four times; disease of the blood, once; circulatory disturbances, twice; bronchiectasis, four times; and indeterminate diffuse interstitial fibrosis, once.

An adequate anatomic diagnosis may be made in the great majority of cases of clinically indeterminate bilateral pulmonary fibrosis, but a specific etiologic factor cannot always be named.

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OXYGEN ADMINISTRATION: AN EMERGENCY METHOD WHEN INTRATRACHEAL TUBE FAILS WITH CHEST OPEN

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THE medical profession is constantly striving for safer operative measures. Treatments are safer if there is preparation for all emergencies. This report is not intended to review the various methods of holding in place intratracheal tubes, nor to describe the endeavor of intratracheal tube designers to prevent an intratracheal tube from slipping out at an inopportune moment. All competent anesthetists strive to avoid accidents of this type. The operating room team should be prepared to meet such an emergency without jeopardy to the patient.

When the patient is being operated upon in the supine position it is usually easy to reintroduce the intratracheal tube if it should accidentally come out. In the lateral position slightly more difficulty is experienced in reintroducing the intratracheal tube. The accidental slipping out of the intratracheal tube is more likely to occur in the prone position of Overholt. In the prone position the reintroduction of the tube is much more difficult. With the chest open there is always reluctance to shift the position of the patient, since gross contamination of the operative field would be the result; even so, this would be better than permitting the patient to become asphyxiated.

After years of open chest operations without the embarrassment of having an intratracheal tube come out, such an accident occurred while operating in the lateral position on an elderly, poor-risk patient with carcinoma of the lung. For a few moments, the anesthetist was concerned with blood administration; during these brief few moments the patient became cyanotic. While the anesthetist endeavored to correct the cyanosis the intratracheal tube came out. It was quickly reinserted but not until the heart had stopped from anoxemia. Cardiac resuscitation was successful after about thirty minutes. The patient lived approximately eighteen hours after operation but did not regain consciousness, even though his color, pulse, respiration. and blood pressure were normal for the major portion of the eighteen-hour period. Had the patient not been cyanotic when the intratracheal tube slipped out it is almost certain that quick reintroduction of the tube would have resulted in little or no embarrassment to the patient. It did not seem possible that such a circumstance would ever happen again, since it had not occurred in all the previous years of experience.

Within one month of this mishap, while operating upon a patient with bronchiectasis in the prone position, the intratracheal tube came out. Before

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it could be reinserted there was cardiac arrest from anoxemia and cardiac resuscitation was not successful. After losing several valuable minutes the thought of introducing oxygen into the bronchial stump occurred, but by the time the necessary simple equipment was assembled it was too late.

In spite of the belief that such accidents would never be repeated, precaution was taken to have available in the operating room an oxygen tank and a sterile rubber tube with a No. 17 intravenous needle attached. Several months later during an open chest operation upon a patient in the prone position the precaution was rewarded when the intratracheal tube accidentally came out.

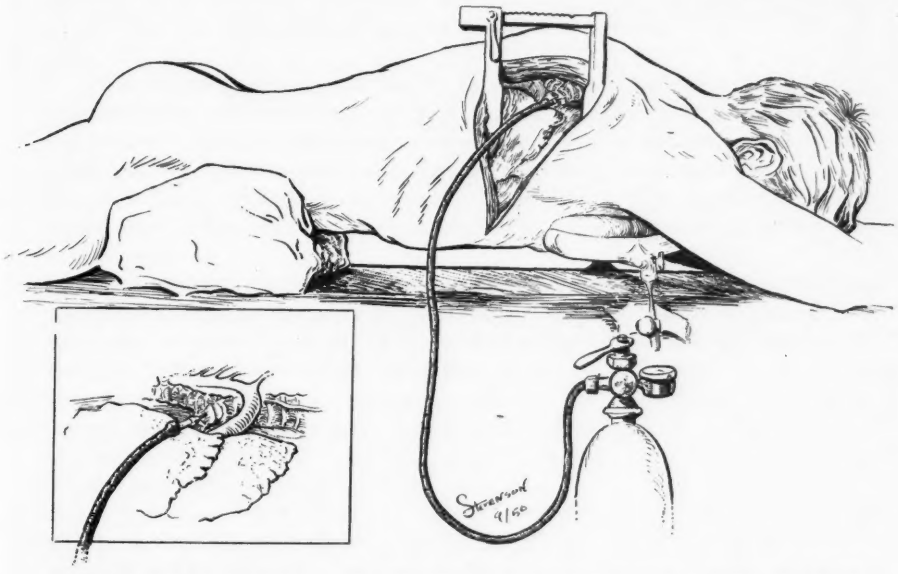

Fig. 1.—The drawing shows the patient in the prone position, and illustrates the simple apparatus necessary to easily and safely introduce oxygen into the tracheobronchial tree should any mishap occur with the intratracheal tube.

Without delay or difficulty the No. 17 needle was inserted into the bronchus and the tubing was attached to the oxygen tank. The assistant held the needle in place and the operator controlled the flow of oxygen from the tank, with the lung under direct vision. When the assistant obstructed the trachea by finger pressure the lung would inflate. Without obstruction to the trachea the flow of oxygen could be increased enough to cause partial inflation of the exposed lung. Apparently the No. 17 needle was a safety mechanism since it would not easily permit an excessive flow of oxygen. The anesthetist reported that the escape of oxygen from the glottis was an aid in reinserting the intratracheal tube. The reinsertion of this tube was done leisurely because there was no change in the heart rate and no sign of asphyxia or anoxemia. The operation was then completed and the patient recovered without further difficulty.

It would, of course, be more desirable to prevent the intratracheal tube from slipping out, but if this should occur it seems obvious that with the chest open one can always insert a needle into the trachea, main bronchus, or lobar bronchus without difficulty. Asphyxia can be avoided by the introduction of oxygen into the tracheobronchial tree. The anesthetist can take ample time to reinsert the intratracheal tube. Overinflation of the lung must be prevented, and apparently it can easily be avoided if a competent person controls the flow of oxygen with the lung in full view. The simple apparatus shown in the drawing (Fig. 1) has proved satisfactory. Obviously this simple apparatus should be included as part of the standard operating room equipment where open chest operations are performed.

BRONCHIAL ADENOMA

Analysis of 26 Cases

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WITHIN recent years there has been an increasing volume of literature dealing with a type of bronchial neoplasm most commonly designated as an "adenoma," but, depending upon the opinion held by various authors concerning its nature and clinical course, described under many other names. Because of the sharp controversy that has arisen as a result of these differences of opinion, an attempt has been made to analyze the series of cases encountered at the Toronto General Hospital in the twelve-year period between 1939 and 1950.

The disagreements engendered by this controversy involve both pathologic interpretation and clinical management. In the first place, many feel that these lesions arise from the epithelium of mucous glands and may be rightly termed adenomas, whereas others[1] have suggested that they arise from maldeveloped bronchial buds—explaining the presence in some cases of both mesodermal and endodermal elements—and thus representing a type of mixed tumor of developmental origin.

In the second place, the disagreements as to clinical management stem from the observations that in many of these cases there have been evidences of local extension, in a few, signs of lymphatic metastases and, recently, reports of occasional hematogenous metastases appearing in the literature, all suggesting that the lesion is essentially malignant in nature. On the other hand, the clinical history is almost always lengthy and there is almost universal operative cure if the lesion can be removed, indicating that the degree of malignancy is minimal. These facts are interpreted by another group of clinicians as meaning that the tumor is benign originally, with a potentiality toward malignant degeneration which may develop late in the course of the disease. On the basis of these two opinions, the argument has been entertained that they may be treated by limited removal instead of the widespread resection favored by others.

With a view to the consolidation of our own opinion on these points, the following analysis was made and is outlined in Tables I to IX below, with a subsequent appraisal and discussion of the views now held, as to the nature of the disease and its proper clinical management. Certain comparisons are made in the clinical course, which seem pertinent in distinguishing it from the frankly malignant bronchiogenic carcinoma, of which a comparison report is now in preparation.

DISCUSSION

It is immediately apparent that the clinical course of these patients parallels very closely the picture presented by previous reports and supports the view expressed by Rabin and Neuhof[2] that there is a very definite "adenoma syndrome" which occurs in a preponderance of the cases. Typically the lesion

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TABLE I. SEX INCIDENCE

	MALE	FEMALE
Adenoma	50	50
Carcinoma	91.1	8.9

TABLE II. AGE INCIDENCE

Average age at onset of symptoms (youngest—21 years) (3 cases) (oldest—64 years)	42.3 years	
Average age at time of operation (youngest—26 years) (2 cases) (oldest—65 years)	47.6 years	cf. carcinoma 56 years
Average length of time between onset of symptoms and operative treatment	5.3 years	

TABLE III. SYMPTOMATOLOGY

	NO. OF CASES	COMPARISON	
		ADENOMA	CARCINOMA
First Symptoms			
Previous lower respiratory infections	9	**34.6**	**16.9**
Cough and sputum	7	26.9	38.7
Hemoptysis	6	**23.1**	**3.2**
Pain (pleuritic)	2	7.7	11.3
Weakness and fatigue	2	7.7	10.5
	26	100.0	
Symptoms present at time of diagnosis			
Cough and sputum (foul odor in 4)	21	80.8	79.8
Hemoptysis (massive in 3)	18	**69.2**	**27.1**
Lower respiratory infection —complicated by empyema in 2 —complicated by bloody pleural effusion in 1	18	**69.2**	**32.3**
Pain—pleuritic type 7 tightness 4 vague discomfort 2	13	50.0	61.3
Loss of weight	12	46.1	58.1
Weakness and fatigue	7	26.9	43.5
Dyspnea	4	15.4	37.1
Wheeze	1	3.8	7.3
Dysphagia	1	3.8	4.0

TABLE IV. RADIOLOGICAL FINDINGS

Atelectasis	18
Consolidation	3
Mass—hilar	6
—peripheral	2
Bronchogram	
Bronchiectasis	2
Complete obstruction of bronchus	2

occurs in either sex with equal frequency and is exhibited in a patient, usually under the age of 50 years, by a long history in which the symptoms of an endobronchial lesion are prominent with emphasis on hemoptysis and the complication of recurring attacks of lower respiratory infection. Physical examination demonstrates the presence of obstructive atelectasis verified radiologically, and in a significantly higher proportion of cases than in carcinoma, the lesion occurs in a major bronchus and may be visualized through the bronchoscope. The sub-

TABLE V. BRONCHOSCOPIC FINDINGS

1. *Positive—Tumor seen* *—Biopsy report adenoma* (Of these 4 were thought to be malignant adenoma due to evidence of infiltration of surrounding tissue and changes in cells.)	18			
2. *Tumor seen—Biopsy report carcinoma* Original biopsy thought malignant and no operation advised. 2 years later lesion in gross resembled adenoma but cells still thought malignant.	*1*	19	73.1	cf. Ca. 57.3
3. *Tumor seen—no definite biopsy report* a) biopsy report granulation tissue (repeated) b) no biopsy taken due to history of hemorrhage c) biopsy report chronic inflammation (possible chondroma). Previous biopsy elsewhere reported as hemangio-endothelioma. d) no biopsy taken as mass thought to be outside left upper lobe bronchus pushing into bronchus.		4		
4. *No tumor seen—confirmatory evidence obtained* —localization of source of bleeding —emergency lobectomy necessary subsequently for continuing hemorrhage	*1*	24	92.3	cf. Ca. 79.8
5. *Negative* a) Lesion in right middle lobe (peripheral mass in X-ray) b) Lesion in lingular bronchus		2	7.7	
		26	100.0	

TABLE VI. DISTRIBUTION OF LESIONS

	RIGHT LUNG	LEFT LUNG	TOTAL
Main bronchus	1	4	5
Upper lobe bronchus	0	1	1
R. intermediate bronchus	7	–	7
R. middle lobe bronchus	3	–	3
L. lingual bronchus	–	1	1
Lower lobe bronchus	5	3	8
Branch bronchus (lower lobe)	1 (dorsal)	0	1
Totals	17	9	26

sequent course suggests benignancy, in that patients in whom a successful resection has been possible, apparently do not succumb to their primary disease.

There are marked differences in emphasis on these features of the syndrome, which make the differentiation from carcinoma feasible on clinical grounds, and the diagnosis should in most cases be suspected before pathologic proof is obtained. Bronchiogenic carcinoma, in contradistinction, tends to occur in men over the age of 50 years in whom the history is relatively short: hemoptysis is less frequent and respiratory infection somewhat less common, particularly as early evidences of the disease. In these cases, a chronic cough often of insidious onset is usually the earliest evidence of the disease, and at a later date the general symptoms such as weakness and fatigue, often associated with dyspnea, appear more prominently in the history. The eventual outcome is of course entirely different, with the majority of patients unfortunately at present dying of their disease unless an early diagnosis is made.

In returning to the controversial points mentioned in the introductory paragraphs, we find it difficult to appraise the pathologic findings in the light of

TABLE VII. PATHOLOGIC FINDINGS

Type of Lesions		
1. Typical adenoma—cells in masses, sheets and cords with tendency to form acini. (In 1 case acini filled with secretion—? cylindroma)		23
2. Mixed tumor type		3
		26
Pathologic Criteria in Benign and Malignant Lesions		
1. Benign adenoma—intrabronchial only		14
—encapsulated		
2. Malignant adenoma		12
a) infiltration surrounding tissues in bronchial wall extrabronchial extension (large extrabronchial mass in only 5) ("iceberg tumor")	9	
b) mitoses (occasional only)	3	
c) variation in cells (slight only)	3	
d) lymph node involvement (both died within 1 month of thoracotomy)	2	
e. Hematogenous metastases (cells found in local blood vessels in 1 case)	0	
		26

the suspected pathogenesis of the tumor. The great majority of the cases fell into the typical adenoma group, being comprised of small round cells, with dark-staining nuclei and little cytoplasm, growing in masses, sheets, and cords with a definite tendency in some cases to form poorly defined acinar spaces. Mitotic figures and variation in size, shape, and staining characteristics of the individual cells were uncommon findings, although a moderate number (approximately one-third of the cases) showed evidences of local extension. Regional lymph node metastases were present in only two cases, and no cases were encountered in which distant blood-stream spread had occurred.

Changes thought possibly to represent instances of the mixed tumor group were found in only three patients. In all three cases the findings were those of heterotopic bone formation with osteoid tissue, and in one case epithelial pearls were present. It was not apparent from a study of these cases whether these changes might have been due to bony metaplasia in islands of cartilage cut off during the local extension of the tumor process, or whether they were actually the result of abnormal developmental processes in the bronchial bud. Consequently, it was found impossible to form any definite opinion regarding the pathogenesis of these two types of endobronchial neoplasm, although it was felt that the entire series could be surveyed as a single group of related cases when considering the question of potential or actual malignancy and the treatment indicated.

On the other hand, fourteen of the twenty-six cases in this series proved to be well encapsulated and showed no sign of change in the constituent cells not in keeping with a benign neoplasm. In addition, the only patient in whom bronchoscopic removal was the definitive form of treatment has survived for nine years and still shows no sign of recurrence. However, there was a relatively high proportion (approximately 20 per cent) of cases showing a large extrabronchial extension—as well as the group mentioned above showing micro-

TABLE VIII. RESULTS OF TREATMENT

TYPE OF TREATMENT	POSTOPERATIVE MORTALITY				POSTOPERATIVE MORBIDITY			
	DESCRIPTION	NUMBER	PER CENT	CARCINOMA	DESCRIPTION	NUMBER	PER CENT	CARCINOMA
1. Lobectomy 11 cases	1 on day after operation (due continuing hemorrhage) 1 due shock on table (hemorrhage) (manipulation)	2	18.2	0		—	0	0
2. Pneumonectomy 11 cases		—	0	10.3	1 empyema (required 3-stage thoracoplasty) 1 empyema (required 3-stage thoracoplasty and local unroofing of residual cavity) 2 cases with spontaneous pneumothorax on opposite side 1 bloody effusion with temp. to 100° F. for 4 weeks	5	45.4	24.1
Resection 22 cases		2	9.1	8.6		5	22.9	22.9
3. Thoracotomy 2 cases	Both thought nonresectable due gross fixation hilus. Removed lymph nodes showed malignant adenoma	2 (within 1 month)						
4. Thoracoplasty 1 case					Persisting discharge from chest sinus, resection not advised due general condition, age and presence empyema			
5. Bronchoscopic Removal 1 case					Symptomless 9 years after treatment			

TABLE IX. SURVIVAL STATISTICS

1. Lobectomy 11 cases	2 operative deaths 9 successful resections none died of disease—all alive AVERAGE SURVIVAL TO DATE (longest—7½ years) (shortest—1 year)	4.3 years
2. Pneumonectomy 11 cases	0 operative deaths 1 still in postoperative period 10 resections followed none died of disease 1 patient lived 4 years and died of carcinoma of sigmoid colon AVERAGE SURVIVAL TO DATE (longest—12 years) (shortest—6 months)	5.4 years
Resections	19 cases followed AVERAGE SURVIVAL TO DATE	4.9 years
3. Thoracotomy 2 cases	Both died within 1 month (glands involved in both)	
4. Thoracoplasty 1 case	Patient alive 6 years later (Duration of symptoms 18 years)	
5. Bronchoscopic removal 1 case	Patient alive 9 years later	

scopic evidence of malignant change—and this alone would, of course, guarantee the failure of any attempt at local endobronchial removal.

It is interesting and important to draw attention at this point to the case of a tracheal adenoma, not included in this series but reported elsewhere,[3] which supports the evidence of others that malignant degeneration may undoubtedly occur in these lesions and often with surprising rapidity, following attempts at local removal by endoscopic means. This patient, a young girl 21 years of age, within two years of endoscopic removal of an apparently benign encapsulated adenoma developed in rapid sequence local recurrence, mucosal infiltration, superficial mucosal ulceration with definite malignant cellular change, followed by infiltration of all the adjacent tissues and organs including the skin.

In addition, there are two cases, also not included in this report, which seem to support the thesis presented by Graham and Womack,[4] that following a long history, these lesions may become frankly malignant, thus obscuring their original nature and making it impossible for the pathologist to distinguish them from the usual bronchiogenic carcinoma. Graham and Womack believe that a change of this type explains the observations that few adenomas were reported prior to the use of the bronchoscope for diagnostic purposes, and that even now the lesion seldom is found at post mortem, whereas carcinoma is discovered with increasing frequency. The first of these patients was a man of 40 years with a definite twenty-six-year history suggesting an endobronchial neoplasm, who was found at operation to have a resectable oat cell carcinoma. Unfortunately, he died at operation as a result of obstruction of the opposite main bronchus by a plug of mucus. There were no evidences of metastasis. The second patient was a woman, 34 years of age, who presented with a seven-year history typical of the adenoma syndrome, and who was found at operation to have a pedunculated endobronchial tumor without microscopic evidence of invasion of the bronchial wall or distant metastases. The lesion proved to be an adenoacanthoma

with irregular glandular formations and masses of cells which were frankly malignant. None the less, this patient is alive two and one-half years after pneumonectomy was performed and completely free of symptoms or signs of recurrent disease.

It would be impossible, obviously, to assert definitely that these two cases of carcinoma arose in pre-existing bronchial adenomas of long standing. It does seem fair to assume, however, that the endobronchial lesions were responsible for the symptoms noted by the patients over such long periods, and, as such, must be accepted as having either developed in originally benign lesions, or else represented from the start malignant lesions of extremely low grade. There would seem to be no doubt that the pathologic process differs fundamentally from the typical bronchiogenic carcinoma with its shorter history and tendency to early metastatic involvement. We prefer to consider that the malignant changes have occurred in a lesion which was microscopically benign at the beginning and which runs a prolonged clinical course that favors this hypothesis.

On the basis of these observations, it is felt that this neoplasm may be rightly termed an adenoma, representing primarily a lesion which is pathologically benign, but which has a definite potentiality toward malignant degeneration. This affects the principles of treatment in such a way that the tumor cannot be treated safely by simple local removal, although it is not necessary to carry out a radical pulmonary resection including the regional lymph nodes with the same objective as in doing a pneumonectomy for bronchiogenic carcinoma. Thus some limited resection of lung would appear to be the operation of choice. With these principles in mind, the following plans for treatment have been utilized, depending upon the stage of the primary disease and the general condition of the patient.

1. *Bronchoscopic removal.*—

Despite the successful result in the one case in which this was attempted, the possibility of a large extrabronchial mass, in addition to the other signs of malignant degeneration, and the instances now recorded of recurrence after apparently complete removal, relegate this procedure to a position of secondary importance in the management of adenoma of the bronchus. This is particularly true in those cases in which extensive irreversible pulmonary disease has occurred distal to an obstructing endobronchial neoplasm, as this can be cured only by resection. It would appear then that bronchoscopic removal should be reserved for those patients who are too ill to undergo an open thoracotomy and possibly as a means of improving drainage of extensive suppuration in the distal lung in order to prepare the patient for subsequent resection of the involved portion of the lung.

2. *Lobectomy or Pneumonectomy.*—

The decision as to the extent of the pulmonary resection indicated in any particular case can be made only at the time of thoracotomy. In the event that there is clinical involvement of the mediastinal lymph nodes, there would seem to be no argument that pneumonectomy is the only treatment of value regardless of the location of the actual tumor. Pneumonectomy also becomes necessary if the adenoma involves the main bronchus, but for lesions located in more distal

bronchi with no evidence of glandular metastases, we feel that lobectomy is the operation of choice. The fact that not a single patient subjected to lobectomy has succumbed to his disease thus far (Table IX) lends considerable support for this opinion. There is no doubt that where this limited resection is possible, better pulmonary function is preserved than in those cases in which a pneumonectomy is necessary.

One cannot overlook the statistical evidence (Table VIII) of a distressingly high incidence of postoperative complications and also the occurrence of operative and postoperative fatalities, which, despite the younger age of these patients, appear more frequently than in similar operative procedures performed in an older group for the more malignant bronchiogenic carcinoma. We believe this is due to the prolonged history which permits more advanced changes to take place in the lung distal to the endobronchial obstruction. Consequently bronchiectasis, pulmonary suppuration, and pleural adhesions are all more marked as complicating factors than in the presence of carcinoma, and, as a result, the resection of the diseased lung cannot be undertaken lightly. The earlier in the course of the disease that diagnosis can be made and treatment initiated, the more likely will one be able to avoid these serious complications.

SUMMARY AND CONCLUSIONS

1. A statistical analysis of twenty-six cases of proved bronchial adenoma is presented in tabular form outlining the symptomatology, the radiological and bronchoscopic findings, and the pathologic attributes of this neoplasm.

2. In similar tabular form, the operative results are presented with an analysis of the operative and postoperative mortality, the postoperative morbidity, and the survival statistics.

3. The clinical findings support the contention that the lesion represents a clinical entity which is associated with a group of symptoms and signs sufficiently definite to be termed an "adenoma syndrome."

4. A review of the pathologic findings with reports of three additional cases pertinent to the discussion suggest that this lesion begins as a pathologically benign neoplasm with a definite potentiality toward malignant degeneration that affects the clinical course and consequently the treatment necessary.

5. On the basis of these observations, it is felt that a limited pulmonary resection of the lobectomy type is the treatment of choice, unless the presence of mediasinal lymph node enlargement or involvement of a main bronchus necessitates a pneumonectomy.

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